Using Multiple Sequential Functional Analysis (MSFA) to identify potential developmental pathways of Non-Epileptic Attack Disorder (NEAD)

Short title: Identifying developmental pathways of NEAD using MSFA

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Thesis abstract

Background. Non-epileptic attack disorder (NEAD) is one of the most common differential diagnoses to epilepsy. Due to the impact of misdiagnosis, research has focused on improving differential diagnosis by identifying factors distinguishing the two populations. These factors, though non-specific and common place comprise much of the understanding of the aetiology of NEAD. Theories which adequately explain the processes by which attacks develop and are maintained are lacking. Although it is agreed that psychological processes underpin NEAD, therapeutic approaches targeting specific processes are underdeveloped. In light of the limitations of currently employed structural approaches, a functional approach may improve understanding of possible mechanisms underpinning NEAD development and maintenance.

Aim. This study aimed to use Multiple Sequential Functional Analysis to explore whether behavioural principles of learning, applied to detailed life histories, can be used to understand the developmental pathway of non-epileptic attacks.

Method. Three adult participants were recruited from outpatient Neurology clinics in the East Midlands, UK. Clinical interviews were conducted using a biographical format to collate detailed information around all aspects of the participant’s histories, current situation, and non-epileptic attacks. To improve the hypotheses made, interview data was triangulated with data from an interview with a relative and a file review. The MSFA was conducted according to the principles of radical behaviourism and applied functional analysis. Data was utilised in the analysis based on the pragmatic truth criterion of functional contextualism.

Results. The results are three detailed functional analytic case studies that track the development of non-epileptic attacks for each participant from formative experiences to their current attack experiences. The results demonstrate that functional analytic principles can be used to understand the developmental pathway of NEAD in these three adults. Though the participants had very different experiences and presentations, an across-case analysis identifies that attacks have similar functional values for these people. Issues including avoiding/reducing stress and emotional suppression appear to be important factors in the development and maintenance of the behaviour.

Discussion. The findings that non-epileptic attacks hold functional value for this group of people, supports the theorised roles of avoidance and secondary gain in the developmental process. The findings have important implications for future research. A strength of the present methodology is that it identifies subtle differences in the learning histories, which has implications for the development of assessment and treatment approaches for those with NEAD.
Acknowledgements

I would like to acknowledge and extend my sincerest thanks to the people that have made this thesis possible:

Dr Mark Gresswell, for his straight-talking support and expertise with the analysis.

Dr Nima Moghaddam, for his invaluable 24/7 responses relating to anything and everything.

Dr David Dawson, for his support with planning the project and analysis, and his ‘gentle’ pressure on the time frame.

Dr Sumeet Singhal for supporting with recruitment and being enthusiastic about the project from the start.

The people who took part in the study, whose time and commitment to share their stories is met with the greatest appreciation.

And finally, my fiancé and my family, for everything.
Statement of Contribution

As the lead thesis researcher I was responsible for the research design, the ethical applications, the collection and analysis of data and the write-up. With regard to the systematic review I was responsible for the review design, searches, the primary assessment of quality and bias and the write-up.

Dr Mark Gresswell (Primary Research Supervisor) contributed to the thesis research design (notably as the co-creator of MSFA) and gave many hours to the functional analyses.

Dr Nima Moghaddam (Research Supervisor) also contributed to the thesis research design, supported with ethical application process issues, and contributed to the functional analyses. With regard to the systematic review Dr Moghaddam second rated the quality and bias of included studies demonstrating inter-rater reliability and finalised the QUOROM flow chart.

Dr David Dawson (Research Supervisor) also contributed to the thesis research design, supported with the implementation of the study, contributed to the functional analyses and reviewed an early draft of the journal article.

Dr Sumeet Singhal (Local Collaborator) advised on the research design and process in relation to his patients, and was responsible for supporting recruitment by providing information sheets to his patients, he also offered advice and support with practical issues including site access and interview venues.
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Systematic Review
Abstract

Background: It is suggested that the communication of a diagnosis of NEAD is the first step in treatment. This suggestion appears largely based on anecdotal reports and a small number of studies which have reported the cessation of non-epileptic attacks after the diagnosis of NEAD is presented.

Objective: The purpose of this systematic review was to examine the impact of receiving a diagnosis of NEAD.

Search strategy: A literature search across the databases Medline, PsycINFO, EMBASE, and CINAHL, and additional hand searching, identified 6 original studies meeting criteria for the review.

Selection Criteria: Included studies were original peer-reviewed articles investigating the impact of receiving a diagnosis of NEAD on adult populations with at least one outcome measured pre and post-diagnosis.

Analysis: The studies were assessed for methodological quality, including biases. This assessment was developed to include criteria specific to research regarding NEAD and diagnosis.

Results: Of the 6 identified studies with a total of 153 NEAD participants, all examined the impact of receiving a diagnosis on seizure frequency. Two examined the impact on health-related quality of life. The findings were inconsistent, with approximately half the participants experiencing seizure reduction or cessation post-diagnosis. Diagnosis appeared to have no significant impact on health-related quality of life. The overall evidence lacked quality, particularly in study design and statistical rigour.

Conclusions: No high quality evidence was found to suggest that receiving a diagnosis of NEAD should be considered a therapeutic intervention. Concerns are considered regarding the appropriateness of seizure frequency as the primary outcome measure and the use of epilepsy control groups. Indications for future research include: measuring more meaningful outcomes, using larger samples and power calculations, and ensuring consistent and standard methods for communicating the diagnosis and recording outcomes.

Keywords: Diagnosis, Non-Epileptic Attack Disorder, Psychogenic Non-Epileptic Seizures, Systematic Review.

Acknowledgements: Review supported by University of Lincoln.
1. Introduction

Non-epileptic attack disorder (NEAD) is the diagnostic term for people who experience non-epileptic attacks [1] which are also commonly referred to as Psychogenic Non-Epileptic Seizures (PNES). There have been many more terms used historically [2], but in this review the terms non-epileptic attacks and NEAD will be adopted. These attacks have been defined as: episodes of altered behaviour which resemble epileptic seizures but are absent of the characteristic clinical and electrographic features of epilepsy [3]. When epilepsy and other medical conditions are ruled out, the attacks are considered to have psychological causes [4]. Although there is no universally accepted theory [5], attacks are widely thought to occur in response to overwhelming distress triggered by difficult situations, thoughts, and emotions [6]. With NEAD clients mainly entering services via the neurology route, the involvement of psychology has been delayed. With growing clinical and academic interest [7], it is anticipated that theoretical understanding and clinical implications will develop.

It has been estimated that 20% - 30% of patients seen in neurology clinics for suspected refractory epileptic seizures are thought to have NEAD [8,9]. Due to the topographical similarities, NEAD is often misdiagnosed as epilepsy, leading to inappropriate and potentially damaging treatment with antiepileptic drugs [10]. It can take an average of seven years before a revised NEAD diagnosis is reached [11]. To remedy this much of the research effort has focused on developing and validating a robust method for the differential diagnosis of NEAD [12]. The method of diagnosis considered the gold standard for sensitivity and specificity involves video-electroencephalogram (V-EEG) monitoring, whereby the electroencephalogram (EEG) records brainwave activity which is considered in conjunction with the clinical characteristics of the seizures observable on the video [13,14]. However, to complicate diagnosis and the identification of appropriate treatment, research using V-EEG data suggests that NEAD is co-morbid in up to 10% of epilepsy patients [15,16]. Research into effective treatments for NEAD has only recently received the attention of systematic reviewers, both concluding that high quality evidence for effective treatments is lacking [17,18].

With comprehensive psychological theories and treatments yet to be established, clinicians often lack a good understanding of NEAD [19]. Consequent inadequate (potentially stigmatising) explanations to the client can lead to confusion, anger, and disagreement with the diagnosis. Such reactions were associated with a poorer prognosis in terms of attack frequency and severity, and quality of life [19]. To provide clinicians with an adequate and non-stigmatising explanation for clients, several protocols have been developed [20,21,22].

Within the literature, receiving a NEAD diagnosis is often referred to as the first stage of treatment [23,24,25]. And rather than this being a figure of speech (as diagnosis is the first stage in most treatment) it appears this is based on the belief that receiving a diagnosis has a therapeutic effect.

Claims that communicating the diagnosis can be considered an intervention in itself largely refer to reports that communicating the diagnosis resulted in the immediate cessation of attacks in some patients, negating the need for further treatment [e.g. 10, 26]. It appears that research has not attempted to explain this phenomenon, or the difference between those whose attacks cease and those whose attacks continue. As with many aspects of NEAD, theory development has fallen short, with categorisation taking its place [27,28]. It has been suggested that three types of NEAD client exist; those whose attacks cease following diagnosis, those whose
attacks reduce/cease following psychological therapy/further intervention, and those whose attacks appear unchanged following diagnosis and therapy [29].

1.1. Rationale
Despite no literature considering the evidence base as a whole, the belief that receiving a diagnosis of NEAD has a therapeutic/intervention effect is commonly held by professionals in the field [23,24,25]. Holding the belief that receiving a diagnosis can reduce/eliminate seizures may lead neurologists to be more considered with their communication of the diagnosis if they see it as a possibly effective therapeutic task. On the other hand, it may perpetuate the historic perception of non-epileptic attacks being considered a factitious/malingering illness [30]. As the role of neurology post-diagnosis is yet to be widely agreed and implemented [31], holding this belief may serve to support services decisions to discharge patients from neurology upon diagnosis and offer no follow-up or formal pathway into psychology services. This lack of agreement is one factor contributing to the slow progress in establishing standard and effective management for clients [32]. When no therapy/post-diagnosis services are available, this belief may be adopted and preferred as a message which can instil hope in professionals and patients. With potential positive and negative implications of holding this belief, it is important to consider the evidence for diagnosis having a positive impact before any conclusions can be made.

1.2. Aims
This review aims to synthesise the evidence regarding the impact of receiving a diagnosis of NEAD. The purpose of this review is to ascertain what the diagnosis impacts on, and whether the evidence is sufficient to draw any specific conclusions regarding the therapeutic effect of diagnosis.

2. Method
2.1. Searching
As previously noted the variation in terminology used in place of non-epileptic attacks and NEAD necessitated a comprehensive and inclusive approach to the literature searching. Also, due to the sparseness of literature in this area, historically used terms now deemed pejorative, such as hysterical seizures, and terms encompassing many phenotypes, such as somatoform disorders, were also included. For searching the databases, groups of terms relevant to two specific elements of the question were combined: terms related to non-epileptic attacks and NEAD; and terms related to diagnosis and outcome.

Electronic searches were as follows:

- CINAHL (1981 to July, week 3, 2014);
- EMBASE (1980 to 2014 Week 29);
- Medline (1947 to July week 3, 2014); and
- PsycINFO (1910 to July week 3, 2014).

Due to issues with differing Boolean operators and truncation of terms the databases were searched separately. The chosen databases include research literature from social science, nursing, and medical professions. The decision to cover this range of disciplines was made due to the changing conceptualisation and continued variation in the management of NEAD.
patients. For full search strategies see supplementary information (online only).

The reference lists of included studies and several relevant reviews [5,38,39] were hand searched to ensure no relevant papers were missed.

2.2. Selection

In order to meet the aims of the review, the authors developed and defined a priori inclusion and exclusion criteria.

Literature was included in the review if it:

- Was original research.
- Included adult participants.
- Explored the impact of receiving a diagnosis of NEAD (or one of its other known terms) with the requirement that seizures with psychogenic non-epileptic origin rather than other medical causes were identified.
- Included one or more outcome measure with data recorded/collected pre and post diagnosis.
- Was written in English (due to the constraints of the study translation was not possible).

Literature was excluded from the review if it:

- Did not specify that the diagnosis was the only ‘intervention’ before outcome data was collected, or if active treatment/intervention was reported following the delivery of the diagnosis and before follow-up data was collected.
- Was not published in a peer-reviewed journal.
- Was not an article length representation of the study (required to assess quality).

A total of 8,011 articles were identified from the electronic searches. The first author reviewed the titles and abstracts of articles for relevance. Articles were excluded at this stage for obvious violations of the inclusion criteria including: unrelated subject matter, papers other than original research and research with non-NEAD populations e.g. other somatoform disorder types. 196 papers remained after this process, with duplicates removed 144 articles remained.

Some articles remained due to the information in the abstract not allowing suitability to be determined, or because no abstract was immediately accessible. Four publications were found to be conference abstracts and were therefore excluded. The authors reviewed full texts for the remaining 140 articles to determine eligibility. Further papers were excluded for the same obvious violations of inclusion criteria. Other reasons for research being excluded included: active treatment before follow-up, presence of treatment not specified, retrospective data collection, and baseline data collected post-diagnosis.
Hand searching of the six included studies [26,33,34,35,36,37] and relevant reviews [5,38,39] identified 12 additional potential studies, with three remaining after the initial abstract sift. Of these, one was a conference abstract and two were excluded when the full-text was reviewed.

2.3 Summary of search and selection process

3. Results

3.1 Data abstraction

General characteristics were abstracted from the six included studies, including: publication year, sample size, study design, outcomes measured, and method of analysis. Additional characteristics relating to the sample were also recorded, including: gender, age, and control group size and
demographics (if applicable). Finally, the findings of each study were abstracted and summarised. All abstracted data are detailed in Table 1.

3.2. Outcomes measured

3.2.1. Seizure frequency

Seizure frequency was measured in all of the studies but included a variety of methods of measuring/recording frequency. Three of the six studies recorded frequency of seizures in numerical form [26,35,36]. Three of the studies used a ranking system of seizure frequency (e.g. none, rare, or regular; monthly, weekly, or daily) [33,34,37]. The method of recording was less clear post-diagnosis; with most studies reporting whether seizure frequency had ceased fully, increased, decreased, or remained the same.

3.2.2. Health-related Quality of Life

Health-related quality of life was measured in two of the six studies [35,37], both using Quality of Life In Epilepsy inventories, QOLIE-31 and QOLIE-10 [40,41]. The QOLIE-31 is a measure of life satisfaction specific to patients with seizures although not specifically non-epileptic seizures. Scores range from 15-100 with a higher overall score representing better health-related quality of life. Within the measure are seven subscales: seizure worry, overall quality of life, energy/fatigue, emotional well-being, cognitive functioning, social life, and medication effects. Psychometric testing using a sample of 304 adults with epilepsy found the lowest internal consistency on the social functioning subscale (0.77) and the highest on the cognitive functioning subscale (0.85) [40]. The QOLIE-10 was found to be highly correlated with the QOLIE-31 and it was concluded that it could be used as a time saving alternative [41].

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1 Seizure frequency will be the term used when reporting directly on reviewed studies, this is to ensure reporting accuracy and also due to the use of epilepsy control groups in some of the studies.
<table>
<thead>
<tr>
<th>Primary author</th>
<th>Publication year</th>
<th>Sample with NEAD</th>
<th>Control group</th>
<th>Methodology</th>
<th>Outcomes measured</th>
<th>Data collection points</th>
<th>Key findings</th>
</tr>
</thead>
<tbody>
<tr>
<td>Duncan</td>
<td>2011 [33]</td>
<td>54 44F, 10M</td>
<td>None</td>
<td>Quantitative Prospective audit Inferential statistics</td>
<td>Seizure frequency</td>
<td>Baseline (pre), 3months (post), and 6 months (post)</td>
<td>24/54 (44%) immediate cessation post diagnosis.</td>
</tr>
<tr>
<td>Farias</td>
<td>2003 [26]</td>
<td>22 14F, 8M</td>
<td>None</td>
<td>Quantitative Repeated measures Inferential statistics</td>
<td>Seizure frequency</td>
<td>24 hours either side of diagnosis</td>
<td>21/22 (95%) reduced including 18/22 (82%) total cessation, 3/22 (13%) 50% reduction.</td>
</tr>
<tr>
<td>Scheepers</td>
<td>1994 [34]</td>
<td>27 20F, 7M</td>
<td>None</td>
<td>Quantitative Retrospective audit Descriptive statistics</td>
<td>Seizure frequency</td>
<td>Pre and post diagnosis</td>
<td>12/27 (44%) increase in frequency, 15/27 (56%) reduction or same frequency.</td>
</tr>
<tr>
<td>Thompson</td>
<td>2013 [35]</td>
<td>19 11F, 8M</td>
<td>NR (sample of 19 split), NEAD, NR</td>
<td>Quantitative RC pilot Inferential statistics</td>
<td>HRQoL, Seizure frequency and intensity</td>
<td>Baseline (pre) and 6-8 weeks (post)</td>
<td>No significant differences in seizure frequency or HRQoL pre and post, or between intervention and control group.</td>
</tr>
<tr>
<td>Wyllie</td>
<td>1991 [36]</td>
<td>20 17F, 3M</td>
<td>Comparison group of 18 children</td>
<td>Quantitative Repeated measures Inferential statistics</td>
<td>Seizure frequency</td>
<td>Baseline (pre) and 1 year, 2 year and 3 year (post)</td>
<td>4/20 (20%) immediate cessation post diagnosis.</td>
</tr>
<tr>
<td>Zhang</td>
<td>2009 [37]</td>
<td>11 8F, 3M</td>
<td>41, ES, 22F 19M, 39 (28-50)</td>
<td>Quantitative Repeated measures Inferential statistics</td>
<td>HRQoL and Seizure frequency</td>
<td>Baseline (pre) and 6-16 months (post)</td>
<td>Improvements in HRQoL but not statistically significant. Significant reductions in seizure frequency.</td>
</tr>
</tbody>
</table>

Notes – F, female; M, male; NR, not reported; ES, epileptic seizures; RC, randomised control; * inferential statistics were used in the analysis but not for seizure frequency related to impact of diagnosis; HRQoL, health-related quality of life
3.3. Key findings

3.3.1. Impact of diagnosis on seizure frequency

All of the reviewed studies provided data regarding the effect of receiving a NEAD diagnosis on seizure frequency. Of the three studies where the primary aim was not to investigate the impact on diagnosis [33,35,36], two reported levels of seizure cessation post-diagnosis [33,36]. Mixed results were reported with seizure cessation in 24/54 participants (44%) in one study [33] and 4/20 (20%) in the other [36]. The third study [35] which primarily aimed to assess the impact of a brief educational intervention on engagement with further treatment, used a diagnosis only control group and reported no significant difference in seizure frequency post diagnosis.

Of the two studies with epilepsy control groups, one reported a significant reduction in seizure frequency in the NEAD and epilepsy control group [37]. Whereas the other [26] reported no change in seizure frequency in the epilepsy control group and a significant reduction in the NEAD group. Specifically, seizures reduced in 21/22 participants (95%), with complete cessation in 18 (82%) and a 50% reduction in seizure frequency for the remaining 3 (13%). It was not reported whether the seizures increased or remained the same in the final participant.

In the final study, which retrospectively reviewed the case notes of NEAD patients [34], it was reported that in 12/27 patients (44%) seizure frequency increased post diagnosis and in the other 15 patients (56%) seizure frequency stayed the same or decreased. However, this study included 15 patients with co-morbid epilepsy and NEAD and did not differentiate the seizure frequency changes in these patients and those with only NEAD.

3.3.2. Impact of diagnosis on health-related quality of life

Of the two studies which investigated the impact of diagnosis on health-related quality of life [35,37], both found no significant difference (positively or negatively) in quality of life from pre- to post-diagnosis. Hypotheses as to why this was the case are considered in the following sections.

3.4. Assessment of Methodological Quality

A meta-analysis was deemed inappropriate for combining and contrasting the results of the studies due to the heterogeneity of the measurement of seizure frequency [42]. Also, as will be later discussed, the quality of the studies raises a concern that an average result across the studies would not be meaningful. Instead, a narrative framework is used to describe the similarities and differences of findings, in terms of the impact of receiving a diagnosis.

It appears that in this area there has been a reliance on certain research to draw conclusions about the impact of receiving a diagnosis of NEAD [10,26]. This may be due to the lack of research, and as was found in this review, investigating the impact of diagnosis was not the primary aim in half of the
studies identified [33,35,36]. Without the systematic method these studies may not have been identified.

In situations such as this it is essential to assess the quality of the relevant research to allow conclusions to be made. Many standardised tools have been produced to assess the methodological quality of research [43]. This has even extended to the development of tools to assess the quality of reviews [44]. However, many of these tools were developed to assess the quality of randomised controlled trials and other specific research designs [45,46] and there is no consensus on which is the best tool [42]. For these reasons and the specific potential quality issues in this area of research, namely varying diagnostic methods, a domain-based quality evaluation tool was specifically developed for this review (see supplementary materials, online only). The developed tool incorporated elements of the Critical Appraisal Skills Programme (CASP) [47] and also considered previous (systematic) reviews relevant to NEAD populations [17,18].

The use of arbitrary cut-off scores in quality assessment tools have been criticised as important quality elements can be masked by the overall score and related overall quality label [48]. Also, single elements of quality can be more important than others in answering posed questions [49]. Therefore, this review adapted the tool developed by the Cochrane Collaboration [42], whereby shades represent levels of quality/bias. Although usually separated within Cochrane reviews, here, quality and bias are combined. No shading signifies low quality/high risk of bias, light shading represents, moderate quality/moderate risk of bias, and dark shading signifies high quality/low risk of bias.

In order to assess the inter-rater reliability of the quality appraisal tool, 50% of the studies (selected at random) were independently rated by two authors (JB and NM). The mean kappa coefficient across items was .75, indicating 'substantial' agreement overall [50].

The individual and synthesised assessment of quality can be seen in Table 3 and Table 4 respectively. Final ratings (presented in Table 3) represent scores agreed between the authors after independent appraisals and discussion of any discrepancies. Table 4 displays the results of the synthesis of the quality and bias of the evidence as a whole. The overall quality and bias is considered by examining how many of the studies were judged as high quality for each criterion.
Table 2. Results of Quality Appraisal

<table>
<thead>
<tr>
<th>Study</th>
<th>Design</th>
<th>Participants</th>
<th>Diagnosis</th>
<th>Outcomes</th>
<th>Statistics</th>
<th>Reporting</th>
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<tr>
<td>Duncan 2011 [33]</td>
<td>+</td>
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<td>N/A</td>
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<td>Farias 2003 [26]</td>
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<td>Scheepers 1994 [34]</td>
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<td>Thompson 2013 [35]</td>
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<td>Wyllie 1991 [36]</td>
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Key

+     Low quality/High risk of bias
++    Moderate quality/Moderate risk of bias
+++   High quality/Low risk of bias
Table 3. Synthesis of quality of evidence

<table>
<thead>
<tr>
<th>Criteria</th>
<th>Overall quality of evidence</th>
</tr>
</thead>
<tbody>
<tr>
<td>1. Power calculation</td>
<td></td>
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<tr>
<td>2. Control group</td>
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<td>3. Demographics</td>
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<td>4. Matched controls</td>
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<td>5. Representative sample</td>
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<td>6. Inclusion and exclusion</td>
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<td>7. Take-up rate</td>
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<td>8. Confounding variables</td>
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<td>9. Attrition rate</td>
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<td>10. Attrition comparison</td>
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<td>11. Diagnostic method</td>
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<tr>
<td>12. Diagnosis delivery</td>
<td></td>
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<tr>
<td>13. Outcome measures</td>
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<td>14. Standardised measures</td>
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<tr>
<td>15. Statistical analysis</td>
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<td>16. Effect size</td>
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<td>17. Reporting bias</td>
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<td></td>
<td>25%</td>
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<td>50%</td>
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<td>75%</td>
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<td>100%</td>
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3.4.1. Results of Quality Assessment

As can be seen in Table 2 the quality between and within the studies is mixed. Five of the studies reported all relevant demographics for the sample but in one study age was not reported [34]. However, in one of the five studies [35] the full sample was split into an experimental and control group but the numbers and demographics of each group were not reported.

Power is the ability of a statistical test to detect a true effect of an intervention (in this case the delivery of the diagnosis) [51]. When adequate power is established the risk of a Type II error is low. Type II errors are false negatives where an effect exists but is not detected [51]. None of the studies reported power calculations. As sample sizes were small it is likely that they do not fully represent all of the population (people with NEAD), and this means the findings are unlikely to approximate population outcomes [51]. Additionally attrition rates may have impacted on the representativeness of the sample in some studies [26,36]. Take-up rates were reported, and found to be high, in four studies [34,35,36,37]; but were not reported in two studies [26,33].

The statistically significant outcomes in the studies suggest that a difference exists but without conducting a meta-analysis it is unclear how robust such findings were to Type I errors. Type I errors are false positives, where an effect is detected but it can be attributed to chance [51]. Additionally, with no studies reporting effect size the magnitude of the differences was unclear [51]. This is particularly important when considering how effective interventions compare to each other [52]. Also, whilst four studies used inferential statistics to calculate the difference between pre and post measures [26,35,36,37], two only used descriptive statistics [33,34]. These were studies in which investigating the outcome of diagnosis was not the primary aim of the research.

Four of the studies accounted for possible confounding variables in their design by using strict inclusion and exclusion criteria [26,33,35,36], one study identified participants with co-morbid ES and NEAD and accounted for this in the design and analysis [37]. One study [34], included clients with co-morbid epilepsy and NEAD and did not differentiate when reporting changes in seizure frequency (which would have been possible due to the study having all individual data available retrospectively).

With regard to NEAD research in particular, five of the six studies [26,33,35,36,37] used the gold standard method for diagnosis, V-EEG monitoring [13,14]. The other study [34] used EEG data without a video overlay which is used to differentiate observable characteristics of seizures [14]. This was also the study which included co-morbid epilepsy and NEAD clients, which is perhaps an artefact of the method of diagnosis used being less specific. The delivery of the NEAD diagnosis was mixed in terms of a clear description but it was made clear in all but one study [33] that participants received the same diagnostic communication. Two of the studies [26,37] reported using a standard well-regarded framework for the communication of the diagnosis [20].
The two studies which used control groups used matched controls of patients who were diagnosed with epilepsy (ES) [26,37].

Seizure frequency was the main outcome and there are no standardised measures for recording this. The two studies which measured quality of life [35,37] used a tool standardised for an epilepsy population, the QOLIE [40, 41]. As previously described, seizure frequency was operationalised differently in the studies. Three were considered to have operationalised the outcome to be measured objectively and clearly [26,33,36], including the two studies where only cessation or continuation of seizures were measured post diagnosis [33,36]. Two studies were considered to use less objective ways of measuring seizure frequency including ranking methods open to bias [34,35]. One study was considered to use a method open to bias and subjectivity which was different at pre and post diagnosis data collection points [37].

Finally, five of the six studies reported in the results and discussion all data/measures described in the method [26,34,35,36,37]. One study collected data which was then not analysed/reported on in the results or discussion [33].

3.4.2. Synthesis of quality

As can be seen in Table 3, the overall evidence is not of a high standard with only four criteria being considered high quality/low bias in over 75% of the studies. The criteria reaching this standard were: take-up rates, reporting of data, diagnostic method, and controlling/adjusting for potential confounding variables. What can be judged and is of particular concern are the two criteria where low quality was identified in all of the studies. Power calculations and reported effect sizes are crucially important in drawing conclusions about presence and magnitude of impact [51]. Therefore the impact of receiving diagnosis may only be minimal and the accuracy of the results suggesting any impact is also questionable.

4. Discussion

This review explored the impact of receiving a diagnosis of NEAD. Six papers were included in this review [26,33,34,35,36,37] to assess the evidence-base and the extent to which receiving a NEAD diagnosis impacts on non-epileptic attack frequency and to a lesser extent health-related quality of life. Results from this review of the literature found inconsistencies in the impact of receiving a diagnosis of NEAD. This may be influenced by the employment of heterogeneous methods of recording non-epileptic attacks. Also the quality of the research in terms of design and statistical rigour was highly questionable. This heterogeneity and lack of appropriate quality makes it difficult for any conclusions to be drawn regarding the impact of a NEAD diagnosis on attack frequency or health-related quality of life.

The claims that receiving a diagnosis of NEAD is a therapeutic intervention were addressed by considering the evidence in this systematic review. This belief, founded on single study findings and anecdotal reports, was not well-supported when the evidence was examined. Health-related quality of life
was measured in two of the six studies, finding no statistically significant changes in pre and post-diagnosis measures [35,37].

Specifically it has been reported that receiving a diagnosis of NEAD can reduce/cease attacks [10,26]. No proposed explanation or theory for why this may occur or why it happens in some people and not others was found in the current literature. Although difficult to calculate due to the heterogeneous methods of recording and reporting, approximately half of participants included in the studies in this review were found to experience a reduction or cessation in non-epileptic attacks post diagnosis. The wide range of reported levels of cessation (20-82%) raises questions about what may moderate response. The inconclusive, variable results and lack of quality found in this review indicate that further research is required.

Without further investigation, it is difficult to conclude whether receiving a diagnosis of NEAD has any impact, positive or negative.

4.1. Limitations

4.1.1. Limitations of this review

The criteria for this review meant that studies must administer measures before and after the diagnosis of NEAD is delivered. This may have led to the exclusion of qualitative research regarding the personal experience and impact of receiving a diagnosis. It is advised that when further research is available which enables conclusions to be made about the objective impact of receiving a NEAD diagnosis, considering qualitative accounts of the impact may support the generation and testing of hypotheses regarding the mechanism of impact.

Strict criteria were imposed on the literature in order to identify studies that would be able to answer the question: What is the impact of receiving a diagnosis of NEAD? Strict criteria applied to a well-researched area would enable the identification of high quality, specific studies which would increase the chance of the question being answered. However, the NEAD research pool remains small and at this time heterogeneous and poor quality studies mean that many questions are yet to be answered. Searching grey literature in any similar reviews in the future may provide more studies for consideration.

It is important to note that numerous papers were excluded as they reported collecting baseline data immediately after the diagnosis of NEAD was delivered (see Table 1.). It was felt that these studies would not answer the question regarding the impact of receiving a diagnosis, although these studies would be included if longer-term follow up post-diagnosis (without active treatment) was investigated. Also, numerous studies were excluded if there was no definitive statement of whether active treatment was implemented prior to follow-up data being collected (see Table 1.). This may be an error in reporting rather than conduct.

With regard to the quality assessment, in this review and generally, many items may reflect assessment of reporting rather than conduct [45]. For example, studies that did not report take-up rates may have been concealing the rates to avoid the criticism of selection bias but may also not report take-up rates as it
used standard service data and all participants who were eligible were included as part of their service/treatment.

The synthesis of the quality assessment was based on the system developed by the Cochrane Collaboration [42] intended to review large amounts of studies. Using this system in the current review with only six studies may be less useful.

4.1.2. Limitations of the studies in the review

The scarcity of research is illustrated by the fact that investigating the impact of diagnosis was not the primary aim in half of the included studies [33,35,36]. Small sample sizes (11-54) were not surprising given the infancy of research into NEAD, it is suggest that the increasing interest in NEAD [7] will enable larger scale research to be undertaken in the future.

Overall the methodological quality of the selected studies was poor. The lack of statistical rigour was apparent across all studies within this review, which limited the ability to draw conclusions. The different methods of recording attack frequency meant that it was difficult to synthesise the findings. The heterogeneity of the studies meant that a meta-analysis to calculate and synthesise effect sizes was not indicated [42].

Although there was consistency with all studies measuring seizure frequency as an outcome, it has been proposed that attack frequency, specifically attack cessation, should not be the primary outcome measured [53]. One study found no significant differences in the employment and benefit status of NEAD participants whose attacks ceased and whose attacks continued. In addition to this, in all participants who remained unemployed (attack free and continued attacks), there was no significant difference in psychopathology (anxiety and depression) [53]. Research has also found that quality of life improved in only 50% of participants whose attacks had ceased [54]. Furthermore, there was no significant correlation between quality of life and attack frequency overall. A study in this review supported the need for further consideration of meaningful outcomes [37]; statistically significant reductions in seizure frequency were found whereas the HRQoL did not show statistically significant improvement. There is also research which has found that cessation of non-epileptic attacks can result in their ‘replacement’ with other ‘conversion’ type symptoms [55]. This limitation can be further generalised to studies of treatment efficacy which also use seizure frequency as the primary outcome measure [17,18,56].

As earlier described, one study did not differentiate between participants with NEAD and participants with co-morbid NEAD and epilepsy, in terms of the post-diagnosis outcome [34]. By not reporting on the outcomes for these groups separately no conclusions can be made about the impact of diagnosis on non-epileptic attack frequency in this study.

4.2. Future research

Future research should endeavour to employ more statistically rigorous designs including larger sample sizes, and calculations of power and effect size. It is possible that research could utilise data already collected as in one of the studies in this review [34]. Twenty years on, data collected during inpatient assessment and diagnosis may be standardised and more comprehensive. This
would enable direct comparisons of pre and post diagnosis measures without the complication of follow-up after discharge increasing the risk of attrition.

It is advised that future research standardises the collection of attack/seizure frequency data using V-EEG monitoring, as in one study included in this review [26]. If due to cost and prioritising equipment for clinical purposes this is not feasible, alternatives such as recording frequency in diaries could be considered. Research should also prioritise measuring psychosocial, psychological, and medical outcomes to further explore their relationship to attack frequency and cessation [37,53,54]. Qualitative research is also indicated in order to explore what clients with NEAD consider a positive outcome.

It would also be favourable for future research to standardise the communication of the diagnosis. Although two studies included in this review [26,37] adhered to a developed communication strategy [20], this has been succeeded by more recently developed protocols [21,22]. With confusion about NEAD associated with poorer prognoses [19], more up to date protocols with more educational information may improve outcomes. It may be useful to compare outcomes after using various communication strategies. A study in this review compared standard diagnostic communication with a brief educational intervention, but found no significant differences in HRQoL or seizure frequency between groups [35].

As noted earlier, the reported levels of cessation in this study were wide ranging (20-82%). If future studies with more rigorous designs continue to find such variability, it would be appropriate to explore what may moderate response. Patient, clinician, or process characteristics (including the communication strategy) may account for variability in the outcomes of diagnosis. Existing research has identified predictors of outcome, typically patient characteristics, but this has focused on responses to active treatment or longer-term follow up rather than diagnosis [10,19,24,36,57,58,59].

Research, including one study in this review, found that epileptic seizures can also reduce post-diagnosis [37,60]. The topographical similarities led to NEAD being commonly misdiagnosed as epilepsy [10]. Although diagnosis has improved, NEAD remains a diagnosis of exclusion with the need for epilepsy to be ruled out [13,14]. This legacy has led to epilepsy patients continuing to be utilised as a control group in NEAD research. With the theory that non-epileptic attacks are underpinned by psychological processes being widely accepted [4], why are patients with epilepsy, underpinned by neurological processes, considered a suitable control group? It may be more appropriate to compare NEAD with other psychological phenomena -further investigation of this is recommended.

5. Conclusions

There is an assumption that receiving a diagnosis of NEAD equates to a therapeutic intervention. The results of this review have found that a limited evidence base of six studies including 153 participants was not consistent or of sufficient quality to draw definitive conclusions regarding this. What can be concluded is that receiving a diagnosis of NEAD should not be presumed to be a therapeutic intervention until we have robust evidence to support this claim.
More rigorous research is required to understand the impact receiving a NEAD diagnosis has on various outcome measures.
References

Supplementary materials

Search strategies

CINAHL search strategy
1. (non#epileptic W2 (attack* or seizure*)) OR (psychogenic W2 (attack* or seizure*)) OR (functional W2 seizure*) OR (hyster* W2 seizure) OR pseudo#seizure* OR (unintended W2 seizure*) or “conversion disorder” or “dissociative disorder” or (dissociative W2 seizure*) OR (non#epileptic attack disorder) OR NEAD OR (psychogenic non#epileptic seizure*) OR PNES OR (psychophysiologic* W2 disorder) or “somatoform disorder” OR “functional neurological disorder*” OR (pseudo#epileptic W2 seizure*) OR convulsion* OR (conversion W2 neurosis) OR pseudo#epilepsy OR (psychogenic W2 symptoms) OR “psychogenic non#epileptic events”.
2. Diagnos* OR post-diagnos* OR outcome* OR prognosis

RESULTS
1: 1,159
2: 625,960
1 & 2: 629
Limited to academic journals = 608
Limited to adult = 223

Post title and abstract sift = 7

EMBASE search strategy
1. Thesaurus terms – pseudoepileptic seizure, psychogenic nonepileptic seizure.
2. (Nonepileptic or non+epileptic) adj2 (attack* or seizure*)
3. (psychogenic adj2 (attack* or seizure*)) or (functional adj2 seizure*) or (hyster* adj2 seizure*) or pseudo#seizure* or (unintended adj2 seizure*)
4. (conversion adj2 disorder*) or (dissociative adj2 disorder*) or (dissociative adj2 seizure*) or nonepileptic attack disorder or non$epileptic attack disorder or NEAD or psychogenic nonepileptic seizure* or psychogenic nonepileptic seizure* or PNES or (psychophysiologic* adj2 disorder) or somatoform disorder*
5. functional neurological disorder* or (pseudoepileptic adj2 seizure*) or (pseudo$epileptic adj2 seizure*) or convulsion* or (conversion adj2 neurosis) or pseudoepilepsy or pseudo$epilepsy or (psychogenic adj2 symptoms) or psychogenic nonepileptic events or psychogenic non$epileptic events
6. Diagnos* OR post-diagnos* OR outcome* OR prognosis

1 = 703
2 = 1650
3 = 1470
4 = 11,189
5 = 33,600
1, 2, 3, 4 or 5 = 45,634
6 = 4,838,059
1, 2, 3, 4 or 5 AND 6 = 15,970
Limited to English Language, journal article, journal or report, EMBASE not medline, and Adult including 65+ = 3802
Post title and abstract sift = 72

Medline search strategy
1. ((nonepileptic or non-epileptic or psychogenic) adj2 (attack$ or seizure$)) OR ((functional or hysteri$ or pseudo or unintended) adj2 seizure$) OR (pseudoseizure$ or pseudo-seizure$) OR (conversion disorder$) OR (dissociative disorder$) OR (dissociative adj2 seizure*) OR (nonepileptic attack disorder) OR NEAD OR PNES OR (psychogenic nonepileptic seizure*) OR (psychogenic non-epileptic seizure*) OR (psychophysiologic* disorder$) OR (somatoform disorder$) OR (functional neurological disorder*) OR (pseudoepileptic adj2 seizure$) OR (pseudo-epileptic adj2 seizure$) OR (convulsion$) OR (conversion adj2 neurosis) OR (pseudoepilepsy or pseudo-epilepsy) OR (psychogenic adj2 symptoms) OR “psychogenic nonepileptic events” OR “psychogenic non-epileptic events”.
2. Diagnos* OR Post-diagnos* OR Outcome* OR Prognosis

RESULTS
1 = 8,037
2 = 4,558,721
1&2 = 4,281
Limited to adult = 2,321
Limited to English language = 1,877

Post title and abstract sift = 50

PsycINFO search strategy
1. (non#epileptic W2 (attack* or seizure*)) OR (psychogenic W2 (attack* or seizure*)) OR (functional W2 seizure*) OR (hyster* W2 seizure*) OR pseudo#seizure* OR (unintended W2 seizure*) or “conversion disorder*” or “dissociative disorder*” or (dissociative W2 seizure*) OR “nonepileptic attack disorder” OR “non#epileptic attack disorder” OR NEAD OR PNES OR (psychogenic non#epileptic seizure*) OR (psychophysiologic* W2 disorder*) OR “somatoform disorder*” OR “functional neurological disorder*” OR (pseudo#epileptic W2 seizure*) OR convulsion* OR (conversion W2 neurosis) OR pseudo#epilepsy OR (psychogenic W2 symptoms) OR “psychogenic non#epileptic events”.
2. Diagnos* OR post-diagnos* OR outcome* OR prognosis

1 = 15,523
2 = 486,992
1 & 2 = 4,906
Limited to academic journals = 4,194
Limited to peer reviewed = 3,998
Limited to adults = 2,109

Post title and abstract sift = 67
## Assessment of Quality and Bias

<table>
<thead>
<tr>
<th>Quality question</th>
<th>Rating Criteria</th>
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<tbody>
<tr>
<td><strong>Design</strong></td>
<td></td>
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<tr>
<td>1 Did the study have adequate power to demonstrate effect?</td>
<td>+++ Power calculations were reported and sufficient; ++ Power calculations were reported but power was insufficient; + There were no reports of power calculations being undertaken.</td>
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<tr>
<td>2 Was there a control group?</td>
<td>+++ There was a control group allowing reasonable conclusions to be made (e.g. epilepsy group also receiving a diagnosis); ++ There was a control group but it only allowed general conclusions (e.g., healthy controls); + There was no control group or their data was not analysed/reported.</td>
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<tr>
<td>3 Are participant demographics reported?</td>
<td>+++ Participant demographics are reported clearly; ++ Participant demographics are reported partially; + Participant demographics are not reported adequately.</td>
</tr>
<tr>
<td>4 If relevant were the groups matched demographically?</td>
<td>+++ Reported demographic variables were matched; ++ Some demographic characteristics were un-matched or data was reported without statistical comparison; + The groups differed in ways that were not statistically corrected or there was no data.</td>
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<tr>
<td>5 Is the sample representative of patients with NEAD?</td>
<td>+++ Sample represents a range of patients with NEAD – appropriate gender split, varied age range and recruited from varied settings; ++ Sample represents a limited range in terms of age, gender or recruitment setting; + Sample has poor representation.</td>
</tr>
<tr>
<td><strong>Participants</strong></td>
<td></td>
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<tr>
<td>6 Are inclusion and exclusion criteria adequately reported?</td>
<td>+++ Inclusion and exclusion criteria are reported clearly; ++ Inclusion and exclusion criteria are reported partially or indirectly; + Inclusion and exclusion criteria are not reported.</td>
</tr>
<tr>
<td>7 Did the study indicate rates and reasons for eligible participants refusing to take part?</td>
<td>+++ Take-up rates recorded and &gt;75%; ++ The study describes take-up rates of between 50 and 75%; + The study describes take-up rates of &lt;50% or take up rates are not reported.</td>
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<tr>
<td>8 Have confounding factors been identified and accounted for in the design and analysis? (i.e. co-morbidity, changes to medication)</td>
<td>+++ Potential confounds are identified and accounted for in the design and analysis; ++ Potential confounds are identified and corrected for in analysis; + Identification of potential confounds are either; only discussed, or completely absent.</td>
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<tr>
<td>9 Is the follow-up of participants complete? Are attrition rates reported?</td>
<td>+++ Evidence of low attrition; ++ Evidence of medium attrition; + No reporting of attrition rates or high attrition.</td>
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<tr>
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<td>Question</td>
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<td>10</td>
<td>Were comparisons made between those lost to follow up and those who participated fully?</td>
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<td>11</td>
<td>Were participants’ diagnosed using V-EEG long-term monitoring the accepted gold standard for diagnostic specificity and sensitivity, accuracy?</td>
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<td>12</td>
<td>Was the diagnosis communicated/delivered in the same way for all participants in the group? Is this clearly described?</td>
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<tr>
<td>13</td>
<td>Were the outcome measures clearly operationalised and matched at pre and post data collection points?</td>
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<tr>
<td>14</td>
<td>Were the measures used standardised?</td>
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<tr>
<td>15</td>
<td>Were appropriate statistical analyses conducted and reported?</td>
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<tr>
<td>16</td>
<td>Were effect sizes adequately reported?</td>
</tr>
<tr>
<td>17</td>
<td>Is all data specified as collected then reported in the results of the study?</td>
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Non-Epileptic Attack Disorder: An examination of the development of non-epileptic attacks using Multiple Sequential Functional Analysis

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WORD COUNT: 7059 excluding references.

Abstract

Background: Non-Epileptic Attack Disorder (NEAD) may affect up to 21,000 adults in the UK and is one of the most common differential diagnosis to epilepsy. NEAD mistaken for epilepsy leads to inappropriate and potentially toxic treatment with medication. For this reason advancing the diagnostic method has been a research priority. In comparison the understanding of the aetiology of NEAD remains limited. A better understanding is required to improve communication, assessment and treatment.

Aim: The present study used Multiple Sequential Functional Analysis (MSFA), an idiographic case formulation method based on behavioural functional analysis, to explore the development of non-epileptic attacks in the histories of three adults.

Method: Data from comprehensive clinical interviews, relative interviews and file reviews were synthesised using MSFA to examine the development and maintenance of non-epileptic attacks across each participant’s life.

Results: Although important differences between participants were identified, all of the attacks appeared to develop from a limited behavioural repertoire in childhood following by an organically underpinned altered state of consciousness with positive consequences. Attacks served to escape aversive experiences and reduce associated stress and in some cases were reinforced by increasing support/care.

Conclusions: MSFA has demonstrated utility in offering a functional understanding of the development of NEAD. Subtle differences between cases have important implications for theory development and treatment planning.

Keywords: Non-Epileptic Attack disorder; Psychogenic Non-Epileptic Seizures; Functional Analysis; Development; Aetiology.

Acknowledgements: Research supported by University of Lincoln and three NHS trusts within the East Midlands.
1. Introduction

Non-epileptic attacks (NEAs) are episodes of altered experience, movement, and/or sensation which resemble epileptic seizures, but are devoid of the ictal electrical discharges in the brain seen in epilepsy [1]. Non-Epileptic Attack Disorder (NEAD) is a diagnostic term for the experience of such events (an alternative term is Psychogenic Non-Epileptic Seizures) [2,3]. Other paroxysmal events including syncope and dystonia could be considered non-epileptic attacks [4], however, the terms non-epileptic attacks and NEAD are typically used by neurologists concerned with attacks considered to be underpinned by psychological processes [5,6].

Incidence has typically been estimated amongst neurology clinic attendees, where between 5-25% of patients seen for suspected refractory epileptic seizures are instead diagnosed with NEAD [7,8,9]. In 2000, using similarly gathered data and epilepsy prevalence in the general population, it was calculated that NEAD may affect between two and 33 individuals per 100,000 of the general population [10].

Early understandings of NEAD were primarily based on observations and case reports. Psychoanalysts proposed that psychic conflict following trauma was converted into physical symptoms in order to reduce anxiety by shielding the conscious self from painful emotions [11]. Behaviourists conceptualised NEAD as a learned behaviour, based on clinical observations that NEAD was commonly seen in people with direct or secondary experience of epilepsy or other altered states of consciousness [12,13]. Indeed, NEAD is co-morbid in up to 10% of people with epilepsy [14,15] and it has been hypothesised that non-epileptic attacks may therefore develop through symptom modelling or observational learning [16]. NEAD was suggested to primarily relieve internal conflict and the support/care elicited by attacks were proposed as secondary gains [17,18].

Once mistaken for epilepsy, it takes an average of seven years for a revised NEAD diagnosis to be made [19]. During this time many people are treated with potentially harmful anti-epileptic drugs [20,21]. Consequently, much of the research effort has focused on identifying risk factors associated with NEAD and not with epilepsy in order to improve differential diagnosis [22]. These factors include: personality disorder [23], specific personality profiles [24,25,26], trauma and childhood abuse [27,28], family dysfunction [29,30,31], and coping strategies including avoidance [32,33].

The identification of differing psychosocial factors in NEAD and epilepsy populations has improved the accuracy of differential diagnosis, for example, by combining a personality profile, with the duration of symptoms and EEG data [34]. However, such psychosocial factors/profiles may present in people with other psychological disorders [29,35,36,37,38]. Additionally, some of these ‘risk factors’ are ubiquitous in the general population, compared to the relative rarity of NEAD [10], for example trauma [39]. Therefore, the proposed risk factors for NEAD appear to be both non-specific and common place, suggesting their presence/absence is only really useful for supporting diagnosis. This critique
calls into question their explanatory utility in understanding the aetiology of NEAD.

Despite this, biopsychosocial and formulation models have attempted to use these factors to understand the development of NEAD [1,40,41,42]. Though individualised formulation is advised due to the suggested heterogeneity of the population [43] understanding any common processes/mechanisms or interactions between factors may inform theory and treatment development.

Current attempts to explain the processes underpinning NEAD include; a pathophysiological mechanism model [44] the integrated cognitive model (ICM) [45-47] and the concept of symptom modelling based on behavioural learning principles [12,13,16]. The pathophysiological mechanism model acknowledges its inability to explain specifically why non-epileptic attacks present and what could be targeted in treatment [44]. The ICM appears to offer a comprehensive explanation for the development of medically unexplained symptoms [45-47]. However, the multitude of treatment targets which have to be distinguished based on individualised formulation [47], indicate it as a meta-model which are suggested to be difficult to verify [48]. [See extended background for a detailed literature review, pg 58]

Despite NEAD being widely conceptualised as a primarily psychological disorder, there has been a general failure in the development of adequate psychological models [22,47]. The limitations of dominant nomothetic structural research suggest that further exploration of NEAD development is necessary, and that a functional approach, based within a specific psychological theory, may be useful for such exploration.

A modern behavioural perspective would consider NEAs to be functional, learned behaviours, maintained by environmental contingencies [49,50]. Functional analysis is the method by which behaviour can be examined in relation to historical learning and consequences [51,52]. Functional analysis has been used to study a wide range of phenomena including: eating disorders, arson, depression and self-harm [53-59].

Multiple Sequential Functional Analysis (MSFA) is a method of functional analysis developed by Gresswell and Hollin [60], to understand complex presentations and the development of behaviour over time. MSFA is a structured, systematic case methodology, underpinned by learning theory principles, which aims to identify the functional development of behaviour over the lifespan. It allows hypotheses regarding the functional relationships between behaviour and consequences to be examined within and across single cases. MSFA has been applied as a research methodology to understand: multiple murder [60], offence paralleling behaviour [61], violent behaviour [62], and most recently female perpetrated intimate partner violence [63]. [See extended background for a full rationale, pg 58]

2. Aims

The current study aimed to undertake a systematic and detailed analysis of three case studies, utilising MSFA, to explore how non-epileptic attacks may
have developed in three individuals diagnosed with NEAD. This will offer a functional understanding of the mechanisms through which NEAD has developed and been maintained. [See extended methodology for research questions, pg 83]

3. Method [See extended methodology for a detailed description of study design, process and analysis, pg 83]

3.1. Ethical approval

The study was granted ethical approval by the host university, an NHS Research and Ethics Committee, and three local NHS trusts.

3.2. Epistemological position

MSFA is grounded in B.F. Skinner’s radical behaviourism [50] and the epistemological position of functional contextualism [64-66]. The primary goal in this philosophy is to predict and influence events with precision in order to construct pragmatic knowledge [64,67,68]. It is this goal and the principles of functional contextualism which guided all aspects of the research process.

3.3. Participants

MSFA is a resource intensive method, requiring multiple interviews from different perspectives over a number of hours, as well as comprehensive file reviews. Consequently, to date published MSFA studies have focused on small samples in order to attempt to capture the processes of interest in depth. For the current study, three participants were recruited.

Participants were recruited from NHS outpatient Neurology services in the East Midlands, UK. A Consultant Neurologist supporting the study disseminated information sheets to his patients (adults) who had a diagnosis of NEAD. Three individuals (one male and two females) contacted the primary researcher and consented to participate, they are referred to as Jayden, Susan, and Daisy (pseudonyms). Table 4 offers demographic information regarding the three participants.

Table 4. Participant demographic information

<table>
<thead>
<tr>
<th>Pseudonym</th>
<th>Jayden</th>
<th>Susan</th>
<th>Daisy</th>
</tr>
</thead>
<tbody>
<tr>
<td>Age (years)</td>
<td>30</td>
<td>62</td>
<td>31</td>
</tr>
<tr>
<td>Age at onset</td>
<td>12</td>
<td>17</td>
<td>29</td>
</tr>
<tr>
<td>Age at diagnosis</td>
<td>24</td>
<td>60</td>
<td>29</td>
</tr>
<tr>
<td>NEAD semiology</td>
<td>Fall followed by prolonged thrashing (akin to tonic-clonic seizure)</td>
<td>Limp limbs (if standing Susan may fall), motionless and unresponsive &lt;1min duration</td>
<td>Falling to become motionless and unresponsive, &lt;1min duration</td>
</tr>
<tr>
<td>NEAD frequency at recruitment</td>
<td>Weekly-monthly</td>
<td>Multiple daily</td>
<td>One in the last 12 months</td>
</tr>
</tbody>
</table>
3.4. Data Collection

With full informed consent from participants, data was collected from three sources. Primary data was collected through extended clinical interviews with each participant, whilst supplementary data for triangulation was collected from interviews with relatives and file review.

3.4.1. Clinical Interview

Clinical interviews were ideographic and focused on obtaining a detailed life history. Details of the participant’s development across all areas of functioning were captured, including childhood, school, friends and intimate relationships, health and work. Suggested risk factors from existing literature, if relevant for the participant, were examined in detail to understand their role (if any) in the functional development of NEAD. The interview style followed the methods of functional analysis assessment and aimed to collate data sufficient to generate a detailed behavioural formulation across the participant’s lifespan [51,52]. Interviews were completed over multiple sessions, lasting between five and seven hours in total for each participant, and were audio-recorded.

3.4.2. Triangulation

Triangulation is typically used to improve validity through cross verification of data from two or more sources [69]. Triangulation was used in this study to gather data pragmatic to the analyses. Discrepancies in the information from the varying sources were resolved through conducting functional analyses. This identified likely influences on the reports/records and enabled the consideration of information chronologically preceding and following the discrepant details.

Relative interviews: The focus of these interviews was influenced by the ongoing functional analyses based on primary data from each participant. Jayden’s mother, Susan’s best friend, and Daisy’s husband were identified as people who had a good knowledge of their histories and their NEAD specifically. Each interview lasted one to two hours and was also audio-recorded.

File review: Documents including letters and session notes relating to relevant incidents (previously identified in the clinical interviews) were noted. Files were accessed from mental health and physical health trusts in the NHS. The relevant notes were considered in relation to primary data and the perspective of relatives.

3.5. MSFA

Functional analysis involves identifying A: B: C: contingency sequences that detail the development and maintenance of a particular behaviour. In an A: B: C: analysis the ‘A’ is the Antecedent; the context which precedes the ‘B’ Behaviours (overt and covert), followed by the ‘C’, the environmental Consequences of the behaviour [51]. The consequences salient in the analysis are those which appear to function to strengthen or reduce the preceding behaviour through the processes of reinforcement or punishment [50]. Table 5 provides a glossary of behavioural terms particularly pertinent to the analyses detailed in this paper.
Table 5. Glossary of behavioural terms

| **Covert behaviour**                          | Internal behaviour, such as cognition, affect, and physiological responses |
| **Overt behaviour**                          | Behaviours which are observed by others                                   |
| **Respondent (classical) conditioning**      | The process by which a neutral stimulus becomes associated with a stimulus which naturally elicits an automatic (reflexive) response (behaviour). The neutral stimulus becomes the conditioned stimulus which can elicit the same behavioural response (conditioned response). |

Once established, conditioned responses (behaviours) can be maintained by operant conditioning

| **Operant conditioning**                     | The process by which behaviours are learnt due to their consequences      |
| **Positive reinforcement**                  | The addition of a stimulus (e.g. consequence) that increases the probability that the preceding behaviour will reoccur |
| **Negative reinforcement**                  | The removal of a stimulus that increases the probability that a behaviour will reoccur |
| **Positive punishment**                     | The addition of a stimulus that decreases the probability that a behaviour will reoccur |
| **Negative punishment**                     | The removal of a stimulus that decreases the probability that a behaviour will reoccur |
| **Generalisation**                          | The process by which the behaviour is elicited by stimulus similar to the original (discriminative) stimulus. |

The term MSFA describes a series of functional analyses across the developmental history of an individual [60]. Whilst typical functional analysis examines discrete behavioural events [51], MSFA seeks to demonstrate the influence of learning on subsequent behaviour development. Within the series, learning based on an A: B: C: sequence at one stage becomes an antecedent or setting event for the subsequent A: B: C: sequence. Following each sequence, these key learning points, hypotheses regarding what seems likely to influence the individual consequent to the detailed events, are proposed. The key learning hypotheses are inferences based on the data collected and the functional analysis developed. The process of MSFA can be demonstrated diagrammatically; with an arrow to represent that key learning in one sequence can become an important antecedent of the next (see Figure 1). As in functional analysis, the order of events in the MSFA implies a demonstration of a functional relationship [70]. The analysis is interested in the consequences which appear to function to strengthen or reduce the specific behaviour (in this case non-epileptic attacks), through operant learning processes. Ramnero and Torneke, offer a particularly accessible overview of such learning processes [71].
Figure 1. The representation of learning in the A: B: C: analyses in MSFA.

3.6. Analysis

Data collection and analysis occurred simultaneously in order to inform further interviews/data collection. The data gathered from the first two clinical interviews was organised chronologically. An initial functional analysis was completed for each key developmental stage in line with agreed procedures for conducting such analyses [51,52,71]. The initially generated MSFA for each participant was used to guide data collection and analysis in the further interviews. Throughout the process a curious stance was taken to ensure equal attention was paid to information which diverged from the developing hypotheses as to confirmatory information. Along with filling any gaps in the history, questions were asked to elicit details surrounding the initial hypotheses. This new information was used to amend the MSFAs and/or add further detail. The amended MSFAs were used to guide the relative interviews and the file reviews, which resulted in further refinements and amendments. This process generated a comprehensive narrative and functional account of the development of NEAD for each participant. For the purposes of this paper the detail in the MSFAs was reduced, however, during this rigorous process the focus was on maintaining the integrity of the narrative and behavioural principles.

4. Results

The three MSFA case formulations are presented below with discussion of functional development at each life stage. These are summaries of salient antecedents, behaviours, consequences and learning in the development of non-epileptic attacks. [see extended results and discussion of analysis for full detailed MSFAs for each participant, pg 97]

4.1. Early experiences

Each participant's formative early experiences are summarised in Table 6. Participants' childhoods were all characterised by limited development of adaptive coping strategies, but due to different circumstances. In terms of trauma Jayden was physically abused, Susan was emotionally abused, and Daisy witnessed significant domestic violence. Notably, for Jayden illness reporting was reinforced as a means of eliciting care, expressing emotion...
appropriately was punished in Susan leading to dissociation, and Daisy was taught to prioritise the needs of others in a 'militant' household.

Table 6. Functional analysis sequence 1: Early experiences

<table>
<thead>
<tr>
<th>Antecedents</th>
<th>Jayden</th>
<th>Susan</th>
<th>Daisy</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>- Jayden is singled out by his father, receiving less toys/gifts than his siblings and being subjected to regular physical abuse</td>
<td>- Susan observes mother punish sister for care seeking and father for emotional expression (both submit to her)</td>
<td>- Daisy’s parents are preoccupied with their issues, conflicts, and portraying a positive public image by spending money</td>
</tr>
<tr>
<td></td>
<td>- Jayden’s mother is unaware of the abuse</td>
<td>- Her parents argue violently at night</td>
<td>- Daisy and her siblings are expected to do chores, and be obedient and quiet</td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>Behaviours</th>
<th>Covert</th>
<th>Covert</th>
<th>Covert</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>- Beliefs of self as unimportant and better off alone</td>
<td>- Beliefs of home as unsafe and expressing emotion as weak</td>
<td>- Beliefs of mother as not good enough (don’t want to be like her)</td>
</tr>
<tr>
<td></td>
<td>- Anger at unfair treatment and not being protected</td>
<td>- Fear of mother and of being close to father</td>
<td>- Feeling valued when productive</td>
</tr>
<tr>
<td></td>
<td>- Dissociation when parents argue</td>
<td>- Dissociation when parents argue</td>
<td></td>
</tr>
</tbody>
</table>

| Overt | - Submit or flight response to violence (avoidance) | - Some expression of emotional distress particularly when ill | - Cares for younger siblings |
|       | - Withdrawal but some comfort seeking (reporting illness to mother) | - Submission to mother | - Completes household chores |
|       | - Seek comfort from sister | - Seek comfort from sister | |

| Consequences | - No positive social relationships are developed | - Emotional expression is positively punished | - Caring is positively reinforced due to relationships developed with siblings |
|              | - Illness increases care from mother (positively reinforced) | - Minimal positive interaction with father | - Caring and doing chores is negatively reinforced by keeping parents happy (reducing conflict) and allows them to work, earning money to buy nice things (positively reinforced) |
|              | - Illness reduces risk of being beaten as mother is present (negatively reinforced) | - Mother causes arguments between Susan and her sister to interrupt their closeness | |
Key learning

- Life is unfair (being singled out/treated worse than siblings).
- Being unwell leads to being comforted.
- Showing negative emotions will be punished
- Even submitting doesn’t improve the situation.
- Others can’t be relied on to be supportive and caring.
- Dissociation can provide relief, being somewhere else rather than here.
- Having nice things/money is important for being happy
- Others’ needs are more important.

4.2. Continued difficulties

In the context of a lack of social skills developed in childhood, Jayden found it difficult to cope with the increased social demands in school and he was verbally bullied. Jayden expressed his anger responding to bullying with physical violence, which was negatively reinforced by reducing the bullying. However, ‘acting out’ at home, swearing and disobeying his mother was punished. Additionally influenced by continued physical abuse from his father Jayden learnt it is best to avoid others. Though continued illness reporting (migraines) elicited care it also enabled him to avoid school. In secondary school Jayden began to play rugby and he was praised for his aggression/anger in this context.

Susan’s mother continued to punish and emotionally neglect her, and she was verbally bullied at school for her appearance. Though they upset her Susan would walk away, hiding that they had hurt her feelings. When the bullying didn’t get worse she saw this as a successful strategy, she feared the bullying would have increased if they had seen her cry (be weak), which is what she had learnt in situations with her mother. Susan spent her time socially with her sister and her friends.

Daisy continued to be pressurised to work hard at school and do chores which she was praised for. She started part-time work from the age of 13 and her wages reinforced working hard. Daisy reported that her younger siblings were treated better than her and given more toys than she was at their age. Daisy felt jealous of them and angry towards her mother, but continued to work hard to buy her own things. She took painkillers for migraines her doctor suggested were stress-related whilst studying for her exams. Daisy completed her A-levels and then began working full-time.

4.3. Organically underpinned altered state of consciousness

Within stressful life circumstances each participant experienced an episode (or in Jayden’s case many episodes) of altered state of consciousness, summarised in Table 7. It is hypothesised that the altered states were elicited automatically (thus considered respondent behaviours). For Jayden the stimulus appeared to be a head-injury resulting in seizures, for Susan over-heating/exertion resulting in fainting, and for Daisy a virus resulting in a blackout/faint. Overall it seems that the episodes had positive consequences, namely avoidance or a reduction in stress.
Table 7. Functional analysis sequence 2: Organically underpinned altered state of consciousness

<table>
<thead>
<tr>
<th>Antecedents</th>
<th>Jayden</th>
<th>Susan</th>
<th>Daisy</th>
</tr>
</thead>
<tbody>
<tr>
<td>Aged 11 Jayden is knocked unconscious during a rugby match and taken to hospital (head injury)</td>
<td>• Frustration and anger at not being able to play anymore (due to head injury)</td>
<td>• I can’t look weak in front of my sister and her friends but I’m not good at this (fear/conflict)</td>
<td>• Aged 20 Daisy becomes unwell with a virus whilst working two jobs, one full-time and an evening job in a pub.</td>
</tr>
<tr>
<td>Jayden is signed off sick from school to recover.</td>
<td>• Jayden experiences post-head injury seizures almost daily</td>
<td>• Susan faints during the race (due to overheating and over breathing related to exertion and emotions)</td>
<td>• Even though I’m stressed and unwell I can’t have time off because I won’t be able to afford the things I want.</td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>Behaviours</th>
<th>Covert</th>
<th>Covert</th>
<th>Covert</th>
</tr>
</thead>
<tbody>
<tr>
<td>Overt</td>
<td>• Jayden experiences post-head injury seizures almost daily</td>
<td>• Susan faints during the race (due to overheating and over breathing related to exertion and emotions)</td>
<td>• Continues to go to work</td>
</tr>
<tr>
<td></td>
<td>• Jayden is diagnosed with epilepsy</td>
<td>• Susan’s sister and friends don’t see her upset (fainting behaviour is negatively reinforced through avoiding feared punishment)</td>
<td>• Daisy is taken to hospital by her boyfriend and has two weeks off work to recover (fainting is negatively reinforced through reducing stress and enabling recovery from the virus)</td>
</tr>
<tr>
<td></td>
<td>• Seizures can cause injuries, and are embarrassing (seizures are aversive)</td>
<td>• Susan’s sister and friends show concern for her well-being (fainting behaviour is positively reinforced).</td>
<td></td>
</tr>
<tr>
<td></td>
<td>• Jayden avoids negative social situations through time off sick (seizure behaviour negatively reinforced)</td>
<td></td>
<td></td>
</tr>
<tr>
<td></td>
<td>• Seizures elicit increased care from mother (seizure behaviour positively reinforced)</td>
<td></td>
<td></td>
</tr>
<tr>
<td></td>
<td>• Mother’s presence reduces father’s violence (seizure behaviour negatively reinforced)</td>
<td></td>
<td></td>
</tr>
<tr>
<td></td>
<td>• Positives derived from rugby stop (seizure behaviour negatively punished)</td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>Consequences</th>
<th></th>
<th></th>
<th></th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>• Jayden is diagnosed with epilepsy</td>
<td>• Susan’s sister and friends don’t see her upset (fainting behaviour is negatively reinforced through avoiding feared punishment)</td>
<td>• Daisy is taken to hospital by her boyfriend and has two weeks off work to recover (fainting is negatively reinforced through reducing stress and enabling recovery from the virus)</td>
</tr>
<tr>
<td></td>
<td>• Seizures can cause injuries, and are embarrassing (seizures are aversive)</td>
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<td></td>
</tr>
<tr>
<td></td>
<td>• Jayden avoids negative social situations through time off sick (seizure behaviour negatively reinforced)</td>
<td></td>
<td></td>
</tr>
<tr>
<td></td>
<td>• Seizures elicit increased care from mother (seizure behaviour positively reinforced)</td>
<td></td>
<td></td>
</tr>
<tr>
<td></td>
<td>• Mother’s presence reduces father’s violence (seizure behaviour negatively reinforced)</td>
<td></td>
<td></td>
</tr>
<tr>
<td></td>
<td>• Positives derived from rugby stop (seizure behaviour negatively punished)</td>
<td></td>
<td></td>
</tr>
</tbody>
</table>
4.4. Onset of NEAD

It is hypothesised that Jayden began to develop non-epileptic attacks as epileptic seizures became better controlled with anti-epileptic medication. This hypothesis is based on different antecedents of seizures/attacks with similar but distinct semiology (the development of NEA antecedents and consequences over time will be explained as the analysis continues). The hypothesised epileptic seizures were triggered by tiredness and photosensitivity, had less pre-ictal aura and were characterised by jerking lasting less than one minute. The hypothesised NEAs initially appeared to be triggered by social demands, were preceded by migraines and were characterised by violent/uncontrolled jerking lasting more than one minute. It is hypothesised that the attack behaviour had become conditioned by the incidental consequences of epileptic seizures; avoiding social demand and increasing care. Therefore due to a severely limited behavioural repertoire, in response to future similar stimuli, NEAs were emitted.

Susan’s successful emotional suppression strategies including walking away from bullies and dissociating at home continued. However, at around 17 years old when Susan was unable to walk away/dissociate in response to becoming upset by work-place bullies it is hypothesised that NEAs mirroring her childhood fainting incident were emitted as the only other strategy in her learning history. It is important to note that this hypothesis is more tentative than others due to less information being collated regarding this time. Susan got married aged 21 and when her husband began to rape and beat her she reported using dissociation to cope. Non-epileptic attacks were emitted in less private situations (mirroring childhood dissociation in the home and the fainting occurring in a social context). NEAs were another strategy for emotional suppression (and thus were negatively reinforced through avoidance of feared punishment), they also elicited increased care and attention from concerned others (they were positively reinforced).

Daisy continued to work hard into adulthood, managing full-time work, keeping her house immaculately clean and caring for her two young children. In response to extreme stress managing her usual duties and planning a birthday party Daisy experienced a spate of severe migraines and an episode weakness in her right side, which was suspected to be a stroke but after an in-patient assessment she was diagnosed with functional hemiparesis. Daisy returned to work part-time on ‘light duties’ after nine months off, despite continued fatigue, pain and migraines (working was reinforced through improved finances and praise). Daisy reported being able to cope with this but her stress and symptoms increased when she was pressurised into resuming her previous supervisory role (which she did because working and putting others’ needs first
had been reinforced in her learning history). As her stress increased, the only behaviour in her history effective in a similar context was the faint. NEAs which mirrored this were emitted and resulted in temporary reductions in stress and symptoms through time off sick (the attacks were negatively reinforced). Though injuries and her children witnessing the attacks were punishing, it is hypothesised that the reinforcement outweighed the punishment as the attacks continued.

4.5. Development (and maintenance) of NEAD

Shortly after onset it appears that Jayden’s NEAs were generalised to occur in response to anger as well as social demands. The events contributing to this generalisation process are outlined in Table 8. It seems that Susan’s NEA continued as a strategy to suppress emotional expression in times of distress. Over time it appears that the positive reinforcement of concern from others reduced as family/friends/colleagues became used to her attacks. They appeared to reduce in frequency when her first husband left her and she began a new relationship with who became her second husband. At a time of increased stress Susan experienced a transient ischaemic attack (TIA) at work. She reported post-TIA symptoms including increased emotionality which appeared to increase her NEAs (as the strategy for suppressing emotional expression). Within the milieu of her symptoms the attacks elicited increased concern from professionals. Daisy continued to function in a reported a zombie-like state. Daisy was advised to quit work and not to drive by her Consultant Neurologist who diagnosed NEAD when attacks continued. Upon quitting work Daisy increased her levels of housework and caring responsibilities, attacks continued. The development of NEAD for each of the participants is summarised in Table 8.

Table 8. Functional analysis sequence 3: Development (and maintenance) of NEAD

<table>
<thead>
<tr>
<th>Antecedents</th>
<th>Key learning from ‘onset of NEAD’ plus…</th>
</tr>
</thead>
<tbody>
<tr>
<td>Father is violent towards Jayden again</td>
<td>People exhibit less concern for Susan following NEAs</td>
</tr>
<tr>
<td>Jayden is not allowed to learn to drive or work</td>
<td>Positive relationship with second husband</td>
</tr>
<tr>
<td>Medication side effects appear to cause weight gain, hair loss and other symptoms</td>
<td>Susan’s children are increasingly demanding of her time and support</td>
</tr>
<tr>
<td>Professionals tell Jayden what to do/not do but nothing improves</td>
<td>Susan is thought to experience a TIA at work</td>
</tr>
<tr>
<td></td>
<td>Daisy is diagnosed with NEAD is told not to drive and is advised to quit work – she is warned that she may end up in hospital again.</td>
</tr>
<tr>
<td>Behaviours</td>
<td>Covert</td>
</tr>
<tr>
<td>------------</td>
<td>--------</td>
</tr>
<tr>
<td>• Anger towards father</td>
<td>• Nothing is ever easy for me for long (anger)</td>
</tr>
<tr>
<td>• Sense of worth developed from increased care and praise for rugby</td>
<td>• I can't cope but I can't let people see me upset (fear of emotional expression)</td>
</tr>
<tr>
<td>• It's not fair (anger and frustration)</td>
<td>• Feeling like this is intolerable (anger/depression)</td>
</tr>
<tr>
<td>• Feeling like this is intolerable (anger/depression)</td>
<td></td>
</tr>
<tr>
<td></td>
<td></td>
</tr>
<tr>
<td>Overt</td>
<td>Overt</td>
</tr>
<tr>
<td>• Expression of anger fighting back against his father</td>
<td>• Non-epileptic attacks increase in frequency</td>
</tr>
<tr>
<td>• Non-epileptic attacks increase in frequency</td>
<td>• Jayden threatens to punch his neurologist</td>
</tr>
<tr>
<td>• Jayden threatens to punch his neurologist</td>
<td>• In a disorientated state Jayden punches people who are trying to rouse him from attacks</td>
</tr>
<tr>
<td>• In a disorientated state Jayden punches people who are trying to rouse him from attacks</td>
<td>• During attacks Jayden damages property when he is jerking/thrashing</td>
</tr>
<tr>
<td>• During attacks Jayden damages property when he is jerking/thrashing</td>
<td></td>
</tr>
<tr>
<td></td>
<td></td>
</tr>
<tr>
<td>Consequences</td>
<td>Covert</td>
</tr>
<tr>
<td>• Jayden’s father has a heart attack and dies (anger punished)</td>
<td>• Continued symptoms including NEAs are investigated</td>
</tr>
<tr>
<td>• Jayden is discharged from the neurology service (anger punished).</td>
<td>• Concern increases (NEAs positively reinforced)</td>
</tr>
<tr>
<td>• Friends and relatives become fearful of being around Jayden</td>
<td>• Susan is signed off sick and eventually quits (NEAs negatively reinforced).</td>
</tr>
<tr>
<td>• Non-epileptic attacks lead to avoidance of feared consequences of expressing anger</td>
<td>• Care from mother increases.</td>
</tr>
<tr>
<td>• Care from mother increases.</td>
<td></td>
</tr>
<tr>
<td></td>
<td></td>
</tr>
<tr>
<td>Key learning</td>
<td>Covert</td>
</tr>
<tr>
<td>• Expressing anger is unsafe</td>
<td>• It is more difficult to cope since the TIA</td>
</tr>
<tr>
<td>• Being around others isn’t safe</td>
<td>• Being stressed seems to lead to NEAs (“blackouts”).</td>
</tr>
<tr>
<td>• Avoiding people and suppressing emotions is best for everyone</td>
<td></td>
</tr>
</tbody>
</table>
4.6. Current context

Table 9 summarises the development of NEAs resulting in their role in the participants’ current context. Jayden lives with his mother and step-father. He has a long-term partner and a three year old daughter. As earlier hypothesised, Jayden continues to have seizures and NEAs, though the seizures are much less common than NEAs. Whilst his NEAs originally seemed to occur in response to social demands, they now also seem to occur in response emotional experiences of anger. Similarly to Susan they act to suppress his emotional expression of anger due to fear of the negative consequences (due to his past learning). In addition to external triggers for anger (typically his partner making demands) NEAs develop to be triggered by his fear of having an attack and the resulting anger that he is in this position. Susan’s NEAs continue to occur to suppress emotions caused by environmental triggers. Susan has become angry that her family support has reduced and that they ask for her support their issues even after her NEAD diagnosis suggested stress as a trigger. Additionally Susan’s NEAs appear to have generalised to occur in response to her internal thoughts about/anticipation of emotionally distressing situations. In the research interviews when Susan talked about past traumatic/emotional events she did not have a NEA, whereas talking about current sources of stress/distress she did. Daisy had a year free of attacks which appeared to relate to her learning new strategies to reduce her pain and fatigue when the punishing value of the attacks increased. Daisy had three attacks in the past six months which appeared to be at times of increased stress (due to financial difficulty) and her increasing her activity levels (with housework and child care).

Table 9. Functional analysis sequence 4: Current context

<table>
<thead>
<tr>
<th>Antecedents</th>
<th>Key learning from sequence 3 plus…</th>
</tr>
</thead>
<tbody>
<tr>
<td>Jayden:</td>
<td>Susan’s family call on her for support</td>
</tr>
<tr>
<td>1. Jayden’s partner makes increasing requests</td>
<td>Susan is faced with stressful situation</td>
</tr>
<tr>
<td>including for him to spend more time with her</td>
<td>e.g. husband becoming ill and benefits being reviewed</td>
</tr>
<tr>
<td>and their child</td>
<td>Susan is asked to talk about current sources of stress in</td>
</tr>
<tr>
<td>2. Jayden being around his daughter</td>
<td>her life (in the interviews)</td>
</tr>
<tr>
<td></td>
<td>Daisy’s children are distressed by her NEAs</td>
</tr>
<tr>
<td></td>
<td>Pain and fatigue continue</td>
</tr>
</tbody>
</table>
Behaviours

Covert
- Anger towards partner.
- It's not safe for me to be around people especially my daughter (fear)
- It's not fair that I can't be alone with my daughter (anger)

Covert
- I can't cope with stress, they should understand this (anger)
- I can't cope with stress, my body can't cope (generalisation to fear of emotional experience)

Covert
- My children shouldn’t be exposed to this (their needs are important)
- I can’t do what I used to, I need to do things differently

Overt
- Withdraw when possible (to mothers house)
- If withdrawal not possible a NEA may occur

Overt
- Non-epileptic attack

Overt
- Daisy goes to bed when her symptoms are bad
- Daisy does less housework

Consequences
- If Jayden withdraws, demands soon continue
- If Jayden has an attack he is left alone to recover (attacks negatively reinforced as they reduce demands and positively reinforced as mother offers care)
- Fear does not reduce

Consequences
- Stress and demands temporarily reduce
- Susan doesn’t have to continue to think about the sources of stress for a short time (NEAs in response to thoughts now conditioned through negative reinforcement)

Consequences
- Daisy’s non-epileptic attacks reduce significantly
- Daisy’s other symptoms reduce (though continue to be debilitating)
- Daisy’s children are less scared
- Therefore doing less is negatively reinforced

Key learning
- Expressing anger is unsafe.
- Being around others isn’t safe.
- Avoiding people and suppressing emotions is best for everyone

Key learning
- Stress should be avoided
- Sometimes there is no trigger, NEAs can happen at any time (Susan does not recognise thoughts as stimuli only external sources)

Key learning
- Self-care is important, it helps everyone.
- Being healthy is more important than being able to buy things and clean the house.

5. Discussion [see extended discussion for further detail, pg 130]

5.1. The development and functional value of NEAD

The analyses presented above suggest that, at least for these three cases, the aetiology and maintenance of NEAD can be understood functionally. All three participants’ NEAs appear to serve to reduce intolerable demands/experiences (external social and subsequently emotional in the case of Jayden, emotional for Susan, and practical for Daisy). Beyond identifying avoidance as a present strategy [32,33] or even proposing it as a common mechanism [11,17], a functional case study approach illustrates subtle differences important in
Further reinforcement for NEAs (or secondary gains) [17,18] of increased care/support appear relevant for Susan and Jayden. These explanations do not indicate that NEAs are conscious or simulated behaviours exhibited in order to achieve intended consequences, though the misconception of behavioural terms may mean it is perceived in such a way [72]. Rather, the development and maintenance of NEAs can be understood using established psychological principles of learning.

The data suggested that childhood experience was key in producing limited behavioural repertoires. Whilst Daisy seemed to have limited opportunity to develop coping strategies, working hard in a controlled environment, Susan’s adaptive coping strategy (expressing distress to seek care) was punished and therefore less adaptive strategies seemed to be inadvertently reinforced. Jayden’s illness reporting behaviour appeared to be the only behaviour effective in eliciting care.

It appears that the behavioural concept of symptom modelling [16] was relevant in the development of NEAD for all participants. However, beyond seizures, other altered states of consciousness (e.g. syncope) were learned and emitted in future similar contexts. Indeed, the semiology of later NEAs (see Table 4.), mirrored the earlier respondent behaviour (seizures or syncope) in each case. Jayden’s NEAs mirror his post-head injury seizures. Susan’s NEAs mirror her incident of syncope (fainting) in childhood. Daisy’s NEAs mirror her incident of syncope (fainting) when unwell aged 20. It appears these altered states of consciousness were relatively unique instances of relief for the participants within difficult/stressful life circumstances. Based on behavioural principles, the MSFAs suggest that when the participants were later in similarly stressful/aversive situations the earlier behaviour was emitted. Within the data gathered regarding their learning histories this seemed to be one of/the only behaviour with ameliorative consequences in terms of escaping aversive situations.

Jayden and Susan’s NEAs appear to generalise as they continued. Jayden’s attacks seemed originally contingent on social demands. Through the process of operant generalisation and in the context of a severely limited behavioural repertoire NEAs appeared to become a response to anger inducing stimuli due to learning negative consequences of expressing anger. Furthermore, he began to fear having NEAs due to learning that others could be hurt by them, making him angry as it restricted him from being around his daughter. As anger was a stimulus for NEAs this appeared to create a cycle serving to confirm his fear but enabling short-term avoidance and a reduction in anger. As Susan’s NEAs continued it appears the reinforced fear of punishment for emotional expression influenced Susan to anticipate emotion inducing stimuli and her thoughts of current sources of negative emotion became a stimulus following which NEAs were emitted.

Conversely, Daisy’s NEA frequency reduced markedly since onset including one year attack-free. The difference between improved and continued NEAD in these cases seems to be that attacks continued when they had positive consequences. Positive short-term consequences are powerful in the context of difficult life experiences even though in the long-term NEAD is not adaptive and
has a negative/restrictive impact [30]. After quitting work and being confronted by her Consultant Neurologist, the punishing consequences of continued attacks (fear of ending up in hospital and the impact on her children) outweighed the short-term relief through resting. Though Daisy was diagnosed soon after onset, a functional explanation suggests that the advice of her Consultant Neurologist was only adopted when the NEA behaviour became ineffective (having more negative than positive consequences). This suggests treatment should focus on modifying behavioural contingencies before reinforcing new adaptive behaviours.

5.2. Relationship to suggested risk factors

Both Jayden and Susan’s NEAs are suggested to be a strategy for suppressing emotional expression. This directly opposes the traditional psychoanalytic concept of conversion: that physical symptoms arise to alleviate (express) emotional pain, related to the memories of childhood trauma/abuse [11].

Susan’s attacks during the clinical interviews exemplify an additional difference; that emotions appear to relate to here-and-now issues. Susan had NEAs during the interviews when discussing current sources of distress, but not when discussing past traumatic experiences. Though this is a nuanced difference, it may challenge the prioritisation of early trauma in explaining NEAD.

Susan having NEAs seemingly in response to talking about current sources of distress and Jayden seeming to have NEAs in response to fear of anger leads to a proposition that experiential avoidance may be a relevant mechanism in NEAD maintenance. Though experiential avoidance (avoidance of thoughts, sensations and emotions in the self) has been proposed as a ‘risk factor’ [73], this is the first time an explanation has been suggested for its higher occurrence in NEAD patients compared to epilepsy and healthy controls.

All participants reported traumatic experiences in their childhoods though none reported childhood sexual abuse. Beyond the presence of trauma as in correlational research [27,28], the functional analyses suggest that these experiences had varying influences on NEAD development. Susan’s early emotional abuse appeared to directly lead to the development of emotional suppression strategies. Though Daisy witnessed significant domestic violence, it appeared to be her upbringing in a strictly controlled family that led her to develop rules about working hard which influenced a lack of coping strategies.

Within these three cases, though Susan’s and Daisy’s attacks manifested similarly, it was Jayden and Susan whose attacks seemed to have functional similarities. Though it is mainly anecdotal literature which suggests semiology can be interpreted, relating to trauma [2,74,75], the MSFAs suggest a different explanation for varying semiology.

5.3. Implications

This exploratory study suggests that MSFA may be able to explain specifically why NEAD develops and produce testable and specific hypotheses for treatment. This addresses suggested limitations with current models [44-47]. The next stage in development would be to seek to verify hypotheses in studies where the MSFA is followed by treatment specifically targeting the hypothesised
mechanisms/processes. Due to ethical concerns regarding the professional support networks of NEAD patients, such studies should be located within services offering such support.

Comparing the three participants’ case conceptualisations, Daisy appears to be a less typical NEAD patient. Though this may spark research interest it is important to consider that in light of Daisy’s improvement, similar individuals may not enter treatment/attract the attention of researchers.

The hypothesised mechanism of experiential avoidance in NEA maintenance (for Jayden and Susan) should be a focus of further intervention research (though this case would be strengthened if identified in further case studies/MSFA research). Acceptance and Commitment Therapy (ACT), is a third-wave behavioural approach which targets experiential avoidance as well as other mechanisms suggested to underpin distress in a range of psychological disorders [76]. Intensive Short-Term Dynamic Psychotherapy (ISTDP) [77-79] underpinned by attachment and psychoanalytic theories, also targets the avoidance of emotions. In both ACT and ISTDP clients with NEAD would be encouraged and supported to connect with, experience and express the emotions and internal experiences they have been avoiding. Given the hypothesised mechanism, it would be likely that clients would have a NEA in the room when discussing/focusing on an emotionally salient issue. The psychologist/therapist would continue this focus when the NEA is over (and the client is safe/unharmed). Behaviourally this would serve to reduce the reinforcing value of NEAs as they would no longer lead to avoidance of the emotion/internal experience. Also, as the client begins to experience and express the emotions in therapy and there are no punishing consequences (as was learned in childhood, hence the development of NEAs as a means of escaping/avoiding emotions), the continued need to avoid and the consequent NEAs will reduce.

5.4. Limitations

A significant limitation of this study was that access to historical files was affected by archiving processes, particularly files for the oldest participant. It may have been that such files could offer more information regarding her early adulthood, when it appears NEAs may have begun. This resulted in less data for the analyses and more tentative hypotheses regarding aspects of the development of NEAD.

This study was developed similarly to a previous application of MSFA [63]. In the previous study the researcher discussed the case conceptualisation with each participant, offering verification of the explanation and assessing acceptability of MSFA as an explanatory framework. Due to requirements of the recruiting service this was not possible in the current study. The developed case conceptualisations were therefore not verified/supported by the participants and their acceptance of a behavioural explanation of NEAD was not ascertained.

6. Conclusions

Structural correlational research and subsequent models have failed to adequately explain the development of NEAD. A functional approach to
understand how suggested risk factors may relate to the mechanisms of NEAD development was indicated. The current study used MSFA to develop hypotheses regarding the functional development of NEAD in three cases. The analyses suggest that NEAD develops from limited behavioural repertoires and the incidence of altered states of consciousness with positive consequences. In line with theoretical understanding NEAs appear to function to reduce aversive experiences through avoidance and appear reinforced by increased care/support. However, subtle differences between cases have important implications for treatment planning. It is suggested that this study has met its aim to offer understanding of the functional development of NEAD, and suggestions have been made for how this informs future research and treatment development.
References


Extended Paper

Background

The background section of this report aims to give an overview of research regarding non-epileptic attack disorder and will consider terminology, incidence/prevalence, diagnosis, semiology, theoretical understanding, suggested risk factors, models and treatment. The rationale for case study research and behavioural case formulation as a research approach will also be detailed.

**Definition, terminology and classification.** Several terms are used interchangeably to describe: observable abrupt episodes of altered behaviour or consciousness, which resemble epileptic attacks but are devoid of the characteristic clinical and electrophysiological features of epilepsy, for which no evidence is found for other organic causes, whereas there is positive evidence/suspicion that psychogenic factors may cause the episodes (Bodde et al., 2009; Cuthill & Espie, 2005; Liske & Forster, 1964). Such terms include: psychogenic non-epileptic seizures (PNES), pseudoseizures, dissociative seizures, dissociative convulsions and non-epileptic attacks (NEAs) (Wiseman & Reuber, 2015).

Including the word ‘seizures’, PNES can be confusing for clients and clinicians, particularly as ‘attacks’ may take the form of absences or unresponsive episodes as in epilepsy *[see Semiology, pg 61]*. Therefore a term not associated with epilepsy such as attacks or events may be preferable (LaFrance & Benbadis, 2006). Conversely, NEAs could technically encompass episodes/behaviour which have an organic aetiology but are non-epileptic, for example, syncope and dystonia (Gates, 2000). However, in a survey of United Kingdom (UK) clinicians, non-epileptic attacks was the most commonly used term (Mayor, Smith & Reuber, 2011). Therefore, for the purposes of this research the terms NEA(s) and Non-Epileptic Attack Disorder (NEAD; Betts & Boden, 1991) were used to describe the previously defined behaviour and diagnosis.
In the late 1800’s and early 1900’s researchers referred to hysteria, or hystero-epilepsy to describe similarly defined behaviour (Breuer & Freud, 1974; Gamgee, 1878, Gomes & Engelhardt, 2013). With the introduction of diagnostic manuals such behaviour was grouped with other somatic symptoms. The *Diagnostic and Statistical Manual of Mental Disorders* (DSM; American Psychiatric Association [APA]) category nomenclature developed from conversion reaction (1st ed.; DSM-I; APA, 1952), to hysterical neurosis (conversion type) (2nd ed.; DSM-II; APA, 1968), to conversion disorder (3rd ed.; DSM-III; APA, 1987). The *International Classification of Disease-10* (ICD-10; World Health Organisation, 1992) groups NEAs within a category of dissociative conversion disorders. These classification systems have and continue to adopt elements of the terms earlier conceptualised by psychoanalysts Freud and Charcot (Allin, Streeruwitz & Curtis, 2005; Owens & Dein, 2006). Terms such as functional seizures currently used by Neurologists and other professionals in the field, are suggested to reflect prevailing aetiological assumptions (Asadi-Pooya & Sperling, 2015). The persistence of terminology linked to psychoanalytic understanding within diagnostic systems suggests a delay in wider understanding, which appears to mirror the delay in theory development [see Early theoretical understanding, pg 63].

NEAD is not included explicitly as a diagnosis within the current DSM, it would instead be categorised as conversion disorder (functional neurological symptom disorder). A diagnosis of conversion disorder is unique in that it explicitly requires exclusion of malingering/feigning. Relatedly, factitious disorder falls within the same category of ‘somatic symptom and related disorders’ (5th ed.; DSM-5; APA, 2013). The association between NEAD (and other somatic symptoms) and malingering is long-standing, Charcot and Freud being the first to propose other explanations in the later 19th century (Breuer & Freud, 1974). The persistence of this association appears to relate to the notion that with no organic cause identified, the symptoms are considered ‘behavioural’, controllable, or purposefully faked for monetary or social gains (Salmon, 2000; Stone et al., 2002). This may be because the veracity of reported symptoms which are not visible is difficult to establish. Additionally, cases of factitious/feigned symptoms (considered to be under voluntary control) have
been verified, using covert video recording for example (Wallace, Sim, Harrison, Bruce & Harbeck-Weber, 2012). However, it is widely agreed that the vast majority of those diagnosed with NEAD do not fake or have control over their attacks (Reuber & Elger, 2003). [see Symptom modelling (learning theory) for a theoretical consideration of volitional behaviour in relation to this research, pg 75]

**Diagnosis.** It can take an average of seven years of living with an epilepsy diagnosis, and related restrictions, before clients receive a revised NEAD diagnosis (Carton, Thompson, & Duncan, 2003; Reuber, Fernandez, Bauer, Helmstaeder, & Elger, 2002). This typically includes years of taking anti-epileptic drugs (AEDs) which present the risk of toxicity and other side-effects (Liske & Forster, 1964; Reuber & Elger, 2003). In fact inappropriate use of AEDs has been associated with increased seizures/attacks (Niedermeyer, Blumer, Holscher & Walker, 1970).

Until relatively recently NEAD was purely a diagnosis of exclusion; using v-EEG data to rule out the presence of epileptic activity preceding, during, and after seizure-like episodes (Mostacci et al., 2011). However, to complicate accurate diagnosis, research suggests that NEAD is co-morbid with epilepsy in up to 10% of people (Benbadis, Agrawal, & Tatum, 2001; Martin et al., 2003).

Experts in the area have more recently collated and presented evidence for signs that distinguish NEAD from epilepsy (LaFrance, Baker, Duncan, Goldstein & Reuber, 2013). Good evidence, sensitivity and specificity was found for the following distinguishing features of NEAD: longer duration, fluctuating course, asynchronous movements, pelvic thrusting, side to side head/body movement, closed eyes, ictal crying, and memory recall. It was advised that these are considered in conjunction with v-EEG data for the most accurate diagnosis to be made.

It is common for those with NEAD not to have an attack whilst under v-EEG monitoring, and as a solution to this induction techniques may be utilised (Leeman, 2009). 39-73% of diagnosing clinicians have reported using induction techniques during monitoring (Schachter, Brown & Rowan, 1996; Stagno & Smith, 1996). These induction techniques utilise placebo agents such as saline...
injections, applying alcohol pads to the body, and head tilting. The most common and controversial technique is that of using placebo injections (Leeman, 2009). Patients are told the injection will induce an attack, enhance movements, or ameliorate movements, with positive responses to the placebo considered criteria for diagnosis (Levy & Jankovic, 1983; Monday & Jankovic, 1993). Following the 2013 guidelines, clinicians may use the induction techniques in order to assess for distinguishing features (LaFrance et al., 2013). Without a better understanding of the mechanisms underpinning NEAD the ability to suggest/induce attacks may strengthen the perception that it is a feigned/simulated disorder.

Carton et al. (2003) found that receiving a NEAD diagnosis can be more distressing when clinicians lack a clear understanding of what NEAD is, and therefore struggle to explain it adequately to clients, something they found to be common. They found an association between confusion, anger and disagreement with a revised NEAD diagnosis and poorer prognosis (in terms of reduction in attack frequency and severity, and quality of life). [see Psychological therapy for NEAD for further consideration of communicating the diagnosis, pg 76]

Semiology. NEAs have been observed to mirror different types of epileptic seizures. “Classic”, or more common, NEAs appear similar to the convulsions seen in tonic-clonic epileptic seizures (Gates, Ramani, Whalen & Loewenson, 1985; Groppel, Kapitany & Baumgartner, 2000; van Merode, De Krom & Knottnerus, 1997). This ‘convulsive’ category of attacks have been described as ‘abreactive’, involving gasping, un-coordinated movement, and characteristic back arching and pelvic thrusting. Conversely, ‘swoons’, as they have been described, are when a patient sinks to the floor and lies inert, flaccid, and unresponsive for a varying amount of time, often with peculiar flickering of the eyelids (Betts & Boden, 1991; Betts & Duffy, 1993). This ‘non-convulsive’ category of attacks appears similar to complex partial seizures (Groppel et al., 2000; van Merode et al. 1997). Associations between gender and attack category have been suggested. Whilst females have been observed to present with convulsive and non-convulsive attacks equally, males have been found to present with convulsive attacks much more frequently than non-convulsive
attacks. Explanations of this observed difference, based on anecdotal reports and associations, concern the acceptability of emotional/behavioural expression between genders and the role of childhood sexual abuse in female convulsive attacks (Betts & Boden, 1991; Betts & Boden, 1992a; van Merode et al. 1997). More recently, a study of manifestations between genders found little difference (Asadi-Pooya, Emami & Emami, 2013).

**Incidence and Prevalence.** Most epidemiological studies have investigated incidence rather than prevalence (Asadi-Pooya & Sperling, 2015). As NEAD is typically mistaken for epilepsy, studies have often been conducted with those presenting to epilepsy clinics and centres. In such clinics NEAD is diagnosed in between five and 25% of cases (Benbadis, O’Neill, Tatum & Heriaud, 2004; Gates, Ramani, Whalen & Loewenson, 1985; Krumholz & Niedermeyer, 1983; Lesser, 1996; Szaflarski, Ficker, Cahill & Privitera, 2000). Based on such data and the prevalence rates of epilepsy, a calculation estimated that NEAD affects between two and 33 people per 100,000 of the United States general population (Benbadis & Hauser, 2000). Though caution is necessary due to the calculation concerning the United States, this estimate suggests that up to 21,318 people in the United Kingdom (UK) may experience non-epileptic attacks (based on the latest population data; Office for National Statistics ONS, 2015).

Some incidence studies specifically concern the outcome of video-electroencephalogram (v-EEG) monitoring to differentiate medically intractable epilepsy (persisting despite the introduction of anti-epileptic drugs; AEDs) and NEAs. For example, in 2002 of the 251 in-patients assessed using v-EEG monitoring 61 (24%) were found to have NEAD (Benbadis, O’Neill, Tatum & Heriaud, 2004). It is unsurprising that the incidence is at the higher end of the range (five-25%) as epilepsy patients whose seizures respond to AEDs would not be seen for inpatient assessment. It is important to note that in this study eight (3%) of those assessed were found to have both epileptic seizures and non-epileptic attacks.

85 (2.2%) of 3781 of new patients presenting to NHS outpatient neurology clinics in Scotland between 2002 and 2004 were initially diagnosed with NEAD.
(Stone et al., 2010). Inclusive of this 85, 587 (16%) were diagnosed with a psychological/functional disorder; the second most common diagnostic category after headaches. This study involved asking Neurologists for their clinical diagnosis following the initial consultation, prior to any formal investigations/tests. The authors reported low diagnostic differences at follow up suggesting accuracy of clinical opinion in diagnosis.

Population based studies estimate annual incidence of NEAD between 1.4 (Iceland; Sigurdardottir & Olafsson, 1998) and 4.9 (Scotland; Duncan, Razvi & Mulhern, 2011) per 100,000. The Icelandic study concerned new cases between 1992 and 1997 and the Scottish study between 2006 and 2008. Using the latter more recent data (which is also perhaps more generalisable in terms of location), an estimated 3,165 new cases of NEAD may be diagnosed each year in the UK (ONS, 2015). Unfortunately the scarcity of comparable research makes it difficult to draw conclusions regarding incidence trends in NEAD.

When NEAD was termed hysteria or hystero-epilepsy it was considered a phenomenon exclusive to females (Massey & McHenry, 1986; Showalter, 1987). Though males are now diagnosed, studies suggest female dominance with an approximate ratio of 3:1 (Alper, Devinsky, Perrine, Vasquez & Luciano, 1995; Bora et al., 2011; Gates, 2002; Krumholz & Niedermeyer, 1983; Reuber et al., 2003). Despite this dominance, Oto, Conway, McGonigal, Russell and Duncan (2005) found that gender made a relatively small contribution to the heterogeneity of the population. This suggests that existing and future knowledge, based on female dominated research samples, can be generalised to males with some confidence. An explanations for this preponderance relates to the higher prevalence of child sexual abuse (CSA) in women and its suggested role in the development of NEAD (Betts & Boden 1991; 1992a). However, this explanation is at the very least too simplistic, given that CSA has been estimated to have a female to male prevalence ratio nearer to 2:1 (Putnam, 2003).

**Early theoretical understanding.** The earliest psychological explanation came from psychoanalytic theory, proposing that psychic conflict resulting from traumatic experiences is converted into physical symptoms to
reduce anxiety and shield the conscious self from painful emotions (see Breuer & Freud, 1974). This appeared based on the observation that clients who presented with non-epileptic attacks frequently reported traumatic histories (Devinsky, 1998).

Though the popularity of behaviourism waned in the 1950’s when it was superseded by the cognitive revolution (Miller, 2003), it was the 1980’s when behavioural explanations for NEAD were proffered as alternatives to the dominant psychoanalytic understanding (Ramani, Quesney, Olson, & Gumnit, 1980). Early behavioural theorists conceptualised non-epileptic attacks as learned behaviour, supported by observations that attacks were mainly found in people with experience (direct or observed) of epilepsy or similar altered states (Hopkins, 1989). In line with the perseveration of psychoanalytic terminology in diagnostic systems, Devinsky (1998) highlighted the influence of psychoanalytic principles on behavioural theory, with relieving internal conflict proposed as a primary gain maintaining NEAD. The behavioural secondary gains described were the support/care elicited by an attack and the avoidance of aversive situations (Devinsky, 1998; McHugh & Slavney, 1998). The suggested secondary gains and the notion that the behaviour is advantageous relate to the description of some NEAD patients adopting a ‘sick role’ (Krawetz et al., 2001; Reuber, 2009).

**The research.** In a review by Bodde et al. (2009), it was found that much of the research literature aimed at identifying the causes of NEAD has focused on correlations of psychosocial factors. Suggested risk factors included: personality traits, trauma (including abuse and neglect), family relationships, emotion recognition and regulation difficulties, and stress and coping strategies. The empirical evidence for NEAD risk factors (including research since the 2009 review by Bodde and colleagues) are summarised below:

**Personality traits.** Personality has been identified as a potential factor in differentiating patients with medically intractable epilepsy and NEAD. Studies focused on personality disorder (PD) and NEAD co-morbidity, have found borderline personality disorder (BPD) in the highest incidences in NEAD patients compared to control groups (typically epilepsy) (see Lacey, Cook &
Salzberg, 2007). Furthermore, having PD was found to be the only predictor of having NEAD in a study of psychiatric disorders in NEAD patients, epilepsy patients and healthy controls (Direk, Kulaksizoglu, Alpay & Gurses, 2012).

Moving away from diagnoses, a considerable research effort has investigated personality profiles in people with NEAD, most commonly using the Minnesota Multiphasic Personality Inventory (MMPI-2) (Butcher, Dahlstrom, Graham, Tellegen & Kreammer, 1989), a 567 item true/false self-report measure. Generally, elevations on clinical scale one ‘Hypochondriasis’ and three ‘Hysteria’ have been found to be suggestive of NEAD rather than epilepsy (Cragar et al., 2003; Derry & McLachlan, 1996; Russell, Coady & Chaytor, 2009). Using the MMPI-2 restructured form (Ben-Porath & Tellgen, 2008) a much shorter measure, Locke and colleagues (2010), found the clinical scale ‘somatic complaints’ most sensitive and specific in differentiating NEAD from epilepsy patients. Binder, Salinsky and Smith (1994) identified different personality profiles in clients with NEAD and clients with epilepsy, which improved diagnostic accuracy from 74% using EEG data, to 81% using EEG data, duration of the symptoms and the profile (Storzbach, Binder, Salinsky, Campbell, & Mueller, 2000). This combination of factors was supported as the most predictive of NEAD patients in a study by Schramke, Valeri, Valeriano and Kelly (2007).

Advocating the move to personality profiles, and in line with the suggested heterogeneity of NEAD patients, Reuber, Pukrop, Bauer, Derfuss and Elger (2004) identified two main and one less common profiles using the Dimension Assessment of Personality Pathology – Basic Questionnaire (DAPP-BQ). Despite their intentions to move from PD diagnoses, the two main profiles actually resembled characteristics of BPD (n=43, approximately 51%) and compulsive PD (n=37, approx. 44%), respectively. Although the other profile was only seen in four individuals (approx. 5%) it resembled the characteristics of avoidant PD. One final participant did not have a profile consistent with any of the others. NEAD patients with BPD profiles/diagnoses have been found to have poorer prognosis in terms of attack cessation (Bowman, 2001; Reuber et al., 2004).
A major limitation of research into discriminative NEAD personality profiles, can be found in the review by Lacey et al. (2007). Only 50% of the studies reviewed (nine out of 18) used control groups, furthermore six of these did not report on the statistical significance of their findings. Additionally, Binzer, Stone and Sharpe (2004) suggest that while such profiles may support diagnosis, they are not distinct to clients with NEAD; rather they reflect differences commonly present in people with other psychologically underpinned somatic phenomena. For example, one study found a statistically similar personality profile in people with NEAD and people with insomnia (Bodde et al., 2011). This suggests such profiles are only useful for supporting the NEAD/epilepsy differential diagnosis process and do not offer anything to improve understanding of the aetiology of NEAD.

It is important to consider the concept of personality and personality pathology in terms of how it may relate to NEAD. Having a personality disorder refers to having enduring, inflexible, and maladaptive patterns of inner experience and behaviour, that deviate markedly from cultural expectations and lead to significant distress or impairment (DSM-5; APA, 2013). By this definition, NEAs could be considered a symptom of a PD. The main proposition however, is that the two are co-morbid diagnoses related by similar formative experiences e.g. trauma (Harden et al., 2009; Lacey et al., 2007). In either case, categorising and treating people based on how they present, in the case of mistaken diagnoses of epilepsy for example, has proved problematic (Reuber & Elger, 2003). Describing what people do, the mainstay of medical models and psychiatry, can direct simplified targets for treatment, e.g. to fix maladaptive patterns. However, as the PD description suggests, the patterns are enduring and inflexible. Thus understanding how the patterns developed and are now maintained is likely to aid understanding and treatment planning (how to ‘fix maladaptive patterns’).

**Trauma.** Many early observations and later correlational studies have led to the emphasis of the role of trauma in the aetiology of NEAD (Arnold & Privitera, 1996; Bowman, 1993; Bowman & Markand, 1996; Nash, 1993; Rosenberg, Rosenberg, Williamson & Wolford, 2000). In a review of 17 studies into trauma and NEAD, Fiszman, Alves-Leon, Nunes, D'Andrea and Figueria
found high rates of lifetime physical and/or sexual abuse between 50% and 77%. Additionally the authors suggested that studies finding lower rates may be due to limiting definitions of abuse, though in opposition extremely broad definitions may explain the higher rates. Conversely, they advise caution with regard to the clinic/hospital samples in the reviewed studies. They note that there will always be higher levels of adverse event experience in such samples compared to community samples, which can lead to spurious associations. Much less focus has been placed on neglect as a form of trauma. One study found 42.4% of NEAD patients compared to 26.7% of epilepsy patients reported experiencing childhood neglect (Akyuz, Kugu, Akyuz & Dogan, 2004).

Sharpe and Faye (2006) conducted a systematic literature review and meta-analysis of the relationship between CSA and NEAD. The review concerned 34 studies published between 1992 and 2004. In 32 studies that distinguished CSA from other forms of abuse, on average 33.2% of participants with NEAD reported experiencing CSA (range: 5.9-84.6%). 19 effect sizes were calculated based on data from 14 studies. With an odds ratio index of 2.940 (95% confidence interval 2.291-3.772), CSA was three times more likely to be reported by those with NEAD than those in the control conditions (typically an epilepsy control group). A limitation of this review is the likely impact of the limited search terms used (non-epileptic and pseudo seizures), particularly as earlier described the terminology for NEAD has and continues to vary. The authors did however highlight the limitation of cross-sectional designs. Most important is their acknowledgement that CSA and other early traumas cannot be isolated from later experiences and related mediating variables to infer a direct causal path.

With regard to mediating factors, Salmon, Al-Marzooqi, Baker and Reilly (2003) found childhood family dysfunction, specifically control in the family (the predominance of rules over individual interests and needs), as a mediator between CSA and NEAD. The authors hypothesised that NEAs might be a way in which an individual learns to exert control in such environments. Family dysfunction as a mediator was also found in a study investigating the relationship between CSA and more general dissociative psychopathology (Nash, Hulsey, Sexton, Harralson & Lambert, 1993). These studies highlight the
importance of wider context in understanding psychosocial factors and the development of NEAD/related psychopathology. However, caution is advised given that the experience of CSA may influence retrospective perceptions of childhood generally (Nash et al., 1993).

**Family relationships.** Further to family dysfunction as a mediator between CSA and NEAD/related psychopathology (Nash et al., 1993; Salmon et al., 2003), research has focused specifically on family relationships and functioning. Moore, Baker, McDade, Chadwick and Brown (1994) used the Family Environment Scale (Moos & Moos, 1981) and found the NEAD group reported lower cohesion and higher conflict scores compared to both an epilepsy group and healthy controls. More recently, Binzer, Stone and Sharpe (2004) found that NEAD patients reported significant differences in parental rearing to patients with epilepsy. The former reported experiencing less emotional warmth and more rejection from both parents. This significant difference suggests that perhaps neglect should be given equal attention as abuse when considering early experiences in NEAD patients (Akyuz et al., 2004).

Griffith, Polles & Griffith (1998) considered the role of family circumstances on NEAs, through interviewing seven families each including a child with NEAD, and seven families each including an adult with NEAD (six females and one male composed the children and adult samples). Two raters identified ‘unspeakable dilemmas’ (family/social/religious/political circumstances imposing forced choices on the patient, leading to suffering and distress, which must remain hidden from those involved) in 13/14 cases. They explained their findings as a demonstration that NEAD can mark a patient's suppression of expressing distress, relating this mainly to hiding experiences of sexual and/or physical abuse. In two cases it was another family member, not the patient, who was at risk of abuse.

With 31 adults with NEAD and an epilepsy control group, Krawetz et al. (2001) found significantly higher levels of relational psychopathology in the NEAD patients’ families. The study used self-report questionnaires (concerning family dynamics) and the same questionnaires were completed by first-degree
relatives, responding to Moore et al.’s (1994) recommendation to explore congruence in perceptions of family functioning. The NEAD group data from the McMaster Family Assessment Device closely resembled data from psychiatric populations and the epilepsy group data resembled nonclinical sample data (Kabakoff, Miller, Bishop, Epstein & Keitner, 1990). The NEAD patients perceived particular deficits in affective involvement, communication, conflict, and general functioning. They agreed with Griffith et al.’s (1998) interpretation that NEAD patients may experience difficult in directly (verbally) articulating their needs and emotions, either due to poor communication skills or a perception that direct communication is not encouraged by the family. They also supported Moore and colleagues’ (1994) finding of deficits in cohesion, as NEAD patients perceived their family to lack interest in and the valuing of the activities of each other. Family members of the NEAD group did not perceive difficulties in the aforementioned areas, instead they expressed significant distress regarding the definition of family roles. From their perspective, the NEAD patient is avoiding certain duties (e.g. parenting), by assuming a sick role. Though the perceptions of NEAD patients and their families are incongruent, family therapy would still be appropriate, if not more so due to the conflicting perceptions.

Following on from evidence that interpersonal difficulties, family dysfunction and early traumatic experiences appear common in NEAD patients, Holman, Kirkby, Duncan and Brown (2008) considered the relationship between attachment, early trauma and NEAD development. Attachment is the propensity of human beings to make strong affectional bonds to caregivers (Bowlby, 1969). Attachment theory has been developed to postulate four adult attachment styles borne out of different infant-caregiver relationships (Bartholomew & Horowitz, 1991; Griffin & Bartholomew, 1994). Holman and colleagues (2008) hypothesised that NEAD patients would be more likely to exhibit insecure attachment styles and report more traumatic childhood events than epilepsy patients. In their sample of 17 NEAD patients a fearful attachment style was significantly more frequent, and significantly more abuse and neglect was reported compared to reports by epilepsy patients. Fearful attachment is suggested to be characterised by a desire to be emotionally close to others but
experiencing difficulty trusting them not to hurt or let you down (Bartholomew & Horowitz, 1991; Griffin & Bartholomew, 1994).

Attachment style, however, was not a unique predictor of NEAD; both early trauma and fearful attachment added significantly to the predictive power (with anxiety and depression controlled for). The authors noted study limitations including small sample size leading to low statistical power, not assessing disorganised attachment (commonly associated with dissociative psychopathology) and that the questionnaire data may offer less information and reveal less difference than interview data regarding abuse (Holman et al., 2008). More generally, fearful attachment has been found to fully mediate the link between childhood trauma and somatisation in a community sample of women (Waldinger, Schulz, Barsky & Ahern, 2006). In this study the relationship in the male sample differed; both childhood trauma and an insecure attachment style contributed individually to predicting somatisation. Though gender made a relatively small contribution to the heterogeneity of the NEAD population (Oto et al., 2005), this may differ in more general somatisation populations.

Emotion related deficits. Emotional regulation is widely agreed to be a product of successful early relationships (attachments), where soothing is modelled by the parent and learnt by the infant. Conversely emotional dysregulation is the poor modulation of emotion due to absent, abusive or inconsistent caregiving (Bartholomew & Horowitz, 1991; Bowlby, 1969; Sroufe, 2005). Uliaszek, Prensky and Baslet (2012) identified two distinct emotional regulation profiles in a self-report study of 55 NEAD patients. One group reported little emotional dysregulation but high levels of avoidance and emotional unawareness. A second group reported significant emotional regulation difficulties in addition to more psychiatric symptoms and lower quality of life. The identification of two distinct subgroups in terms of emotional dysregulation was supported in a similar study, though avoidance was not assessed (Brown et al., 2013). Reuber et al. (2004) found that NEAD patients had significantly higher emotional dysregulation scores on the DAPP-BQ than epileptic and healthy controls (p<0.001).
Alexithymia, the sub-clinical deficit in identifying and describing your own emotions (Sifneos, 1973), was investigated as a potential factor in differential diagnosis. Though alexithymia has been found more commonly in NEAD patients than in epilepsy patients in some studies (Bewley, Murphy, Mallows & Baker, 2005; Kaplan et al. 2013), it has been concluded that alexithymia does not have discriminative utility (Bewley et al., 2005; Myers, Matzner, Lancman & Perrine, 2013; Tojek, Lumley, Barkley, Mahr & Thomas, 2000). Considering alexithymia as a state-dependent phenomenon rather than a fixed trait, though unhelpful in diagnosis, alexithymia and related coping strategies have been hypothesised as appropriate targets for treatment (Bewley et al., 2005; Myers et al., 2013).

**Stress and coping strategies.** Though as previously detailed, NEAD patients may report more trauma and family issues than epilepsy/healthy controls, constituting more stressful life experiences, it has been proposed that the perception of events (attention), rumination, and importantly coping strategies may influence the reporting (Testa, Krauss, Lesser & Brandt, 2012; Tojek et al., 2000). It has been found that NEAD patients report more distress in response to negative life events (Testa et al., 2012; Tojek et al., 2000), and employ fewer practical coping strategies than healthy controls (Testa et al., 2012). A self-report study of 82 NEAD patients found that in stressful situations one third tended to employ less effective emotion oriented strategies, and one quarter under-employ effective task-oriented strategies, compared to adult norms (Myers, Fleming, Lancman, Perrine & Lancman, 2013). A limitation of these studies is that the role of coping strategies in the development of NEAD cannot be ascertained as participants with pre-existing NEAD were reporting on current strategies. Additionally whilst participants reported less adaptive coping than healthy/adult controls, those with other psychological disorders may be similar. In the latter study psychiatric inpatient norms were available for the coping strategy measure, but the researchers compared participant scores to “normal” adult norms basing this choice on being in accordance with typical clinical practice (Myers, Fleming et al., 2013). Evidence that NEAD populations and other psychological disorder populations appear to share similar family functioning experiences (Kabakoff et al., 1990; Krawetz et al., 2001), suggests
that perhaps this profile of coping strategies in NEAD is not unique, using the psychiatric inpatient norms in the above study would have been useful in ascertaining this.

Avoidance was hypothesised by both early psychoanalytic and behavioural theorists as an important factor in NEAD (Breuer & Freud, 1974; Devinsky, 1998). Indeed, self-report evidence suggests that NEAD patients use more avoidance strategies in times of stress and conflict (Frances, Baker & Appleton, 1999; Goldstein, Drew, Mellers, Mitchell-O’Malley & Oakley, 2000; Myers, Fleming et al., 2013), though again in comparison to healthy controls. NEAD patients have been found to have increased basal cortisol levels (Bakvis et al., 2010), associated with stress-sensitivity in the form of attentional bias to angry faces (Bakvis et al., 2009; Bakvis, Spinhoven, & Roelofs, 2009). Based on their previous findings and self-reported higher levels of avoidance, Bakvis, Spinhoven, Zitman and Roelofs (2011) tested their hypothesis that NEAD patients may exhibit threat avoidance behaviour. They found that NEAD patients demonstrated increased avoidance behaviour to social threat cues at baseline compared to healthy controls (this was not a significant difference and the authors highlighted that the small sample limited statistical power).

Conversely, though in line with their previous findings (Bakvis et al., 2009), they found following stress-induction, there was a decrease in avoidance of social threat in NEAD patients. This normalisation was suggested to relate to the presence of an investigator during the task, potentially reducing the emotional value of the experimental social threat cues in comparison.

Focusing on experiential avoidance (avoidance of thoughts, sensations and emotions in the self) of anxiety rather than avoidance of anxiety provoking environmental stimuli, Di Maro et al. (2014) found significantly higher levels in NEAD patients compared to epilepsy patients and healthy controls. Experiential avoidance, or avoidance of introspective experience, has been associated with (or proposed as mediator between stressful life events and) other psychopathology (for a review see Chawla & Ostafin, 2007). This suggests that once again research has not been targeting distinct features or markers of NEAD. Alternatively a more positive view may consider this as evidence that NEAD could be treated similarly to other disorders. Relatively recent theoretical
and experimental developments suggest trans-diagnostic therapies (e.g. Acceptance and Commitment Therapy; Hayes, Strosahl & Wilson, 1999) targeting similar processes, deficits and behaviours, including experiential avoidance, may be a cost effective and outcome effective alternative to population/disorder specific therapies (Hayes, Luoma, Bond, Masuda & Lillis, 2006; Muto & Mitamura, 2011).

**Models/theories of NEAD.** The studies summarised in relation to the psychosocial correlates of NEAD are representative of the majority of research into aetiology. Despite some investigation of predictive factors for NEAD, there is no single coherent theory that is able to consider these to explain the psychological mechanisms through which NEAD develops. This has been proposed as one of the main reasons why NEAD treatment remains so limited. (Bodde et al., 2009; Brown, Syed, Benbadis, LaFrance & Reuber, 2011).

Researchers proposing theories and aetiological models, have described and hypothesised the priority of, and relationships between, suggested psychosocial risk factors for NEAD (e.g. Bowman, 1999; Moore & Baker, 1997). Some also chronologically ordered the factors in relation to NEAD onset (e.g. Bodde et al., 2009; Reuber, Howlett, Khan & Grunewald, 2007). Researchers have also tested ‘models’ by verifying the prevalence or predictive value of chosen factors in NEAD patient samples (e.g. Bodde et al., 2013; Kaplan et al., 2013; Reuber et al., 2007). Baslet, Roiko and Prensky (2010) attempted to explain the heterogeneity of NEAD patients by splitting them into sub-groups based on psychosocial factors distinguishing the groups. Reuber (2009) developed an earlier description of predisposing, precipitating and perpetuating factors (Reuber et al., 2007) into a biopsychosocial aetiological model of NEAD. Despite its title, the multifactorial model describes similar factors to those detailed in this thesis, uniquely however, Reuber incorporated hypotheses regarding specific but varying aetiological factors using case examples.

Though integrative formulation in clinical psychology has been advocated (British Psychological Society, 2011), a clear understanding of the processes and mechanisms underpinning NEAD may not only inform treatment but may also offer improved clarity in communicating to patients. This may be
particularly beneficial as unclear communication has been associated with poorer engagement and poorer prognosis (Carton et al., 2003; Shen, Bowman & Markand, 1990). In line with this some theorists and researchers have proposed more explicitly mechanistic models and theories based within specific schools and orientations:

**A pathophysiological mechanism model.** Baslet’s (2011) model is based on functional neuroimaging evidence in conversion and dissociative disorders related to the observed/researched similarities in NEAD. The model suggests that NEAs are facilitated by an unstable and inflexible cognitive-emotional attention system. Baslet hypothesised that NEAD patients may have dysfunction in the medial prefrontal regions and the anterior cingulate cortex of the brain. These areas are responsible for attention processes and emotional regulation with dysfunction leading to instability, as observed in NEAD patients. Further to suggesting mechanisms of underpinning traits/characteristics, Baslet proposed that dysfunction in these areas and the subsequent altered relationship to other neural systems as the likely explanation for attack behaviour and patient experiences. When such regions integrate with other neural systems responsible for behaviour and sensation, they generate a sense of conscious experience and awareness (Baars, 2002). Conversely lack of integration or an altered relationship between systems may contribute to experiences of altered conscious experience and awareness. Baslet (2011) acknowledged that the final behavioural responses (attacks) have not been studied and suggested them to be pre-wired behavioural tendencies.

**Integrative cognitive model (ICM).** Combining various theoretical concepts previously proposed, Richard Brown developed a cognitive model of medically unexplained symptoms (MUS), inclusive of NEAD (Brown, 2004; Brown, 2006; Brown et al., 2011). This model is based on the premise that conscious experience involves a working model of the environment, generated through interpretation and organisation of sensory information stored in memory. This working model is suggested to trigger behaviour, with routine behaviour being automatically controlled through well-learnt cognition and action programs. This system explains the exhibition of complex behaviour with minimal self-awareness or conscious effort. Novel behaviour, involves more
wilful attentional processes and is therefore perceived as deliberate, effortful and in conscious awareness.

Symptoms (including NEAs) are suggested to arise when low level (routine) attentional processes select rogue representations of the nature of symptoms. These representations are thought to be acquired from earlier personal exposure to symptoms, observing symptoms in others, but also through sociocultural transmission or verbal suggestion. When selective attention is paid to physical sensations, symptom-related information, and/or negative affect, the rogue representations are reactivated in the memory system. Additionally, positive and negative reinforcement (positive consequences) of the reactivations and subsequent NEAs are suggested to contribute to further reactivation. Brown has acknowledged that why some people are less able to inhibit automatic activations than others is an aspect of the model which is underdeveloped (Brown et al., 2011). The model suggests Cognitive Behavioural Therapy (CBT) as an appropriate treatment approach, to target factors that maintain activation of symptom action programmes including: catastrophic misinterpretation, body checking, worry/rumination, reassurance-seeking behaviour and avoidance of feared situations. Improving high-level attentional control over related mental representations is advocated through attention training (Brown 2006). The numerous treatment targets fit the proposition that such models are in many ways meta-models; providing a structure to be developed based on the different factors patients present with, making testing/verification in research difficult (Deary, Chalder & Sharpe, 2007). Though these factors contribute to explanatory mechanisms of medically unexplained symptoms, it may be that (fewer) mechanisms specific to NEAD can be identified which may be more amenable to improving understanding as well verification.

**Symptom modelling (learning theory).** Bandura’s social learning theory applied to NEAD, postulates that like other behaviours, NEAD is learnt by the process of modelling (Bandura, 1971). The concept of symptom modelling (Bautista, Gonzales-Salazar & Ochoa, 2008) suggests that people who witness or experience seizures learn the contexts in which the behaviour has positive consequences (Hopkins, 1989; Ramani et al., 1980). This would
explain co-morbidity (Benbadis et al., 2011; Martin et al., 2003), and the association between family history of epilepsy and NEAD (Hopkins, 1989).

The belief that NEAD is factitious may be exacerbated by misconceptions about behaviourism and theories of learning. The term ‘behavioural’ is often taken to suggest that something is within a person’s control; that they have made a deliberate choice to behave in this way (Salmon, 2000; Stone et al., 2002). This contradicts Skinner’s hard determinist stance that free will does not exist, that our behaviour is the result of our evolutionary and environmental history (Skinner, 1971). Though the vast majority of NEAD patients are not perceived to be simulating/feigning or even in conscious control of their attacks (Reuber & Elger, 2003) behavioural theory can explain perceived simulated/factitious symptoms. For example, video evidence where symptoms only occur in the presence of others and cease when the person is alone (Wallace et al., 2012), are suggested to be due to the behaviour being contingent on the presence of a potential carer (this is known as a discriminative stimulus). Considering the person’s learning history, they would have been deprived of care around the time the behaviour was established, and increased care only when others were present would have reinforced the attack behaviour (see Michael, 1993; Wooley, Blackwell & Winget, 1978). It is perhaps, the pervasive belief in free will (Rakos, 2004) and the persistence of mind/body dualism that means NEAD sits uncomfortably with patients and professionals (Salmon, 2007). It is possible that the preponderance of psychoanalytic explanations concerning unconscious conversion may relate to the misunderstanding of behavioural theory and resulting discomfort in professionals and the perception of patient reactions.

**Psychological therapy for NEAD.** As the current study focused on understanding the development of NEAD, the evidence for psychological therapies with NEAD patients is only summarised here. Therapeutic approaches and intervention research are later considered with regard to the study findings [see Implications for research and practice, pg 140].

Regardless of the therapeutic approach, sensitive and considered communication of the diagnosis and psychoeducation have been advocated (Duncan, 2010; Shen, Bowman & Markand, 1990). Often considered the first
stage in treatment (Alsaadi & Marquez, 2005; LaFrance et al., 2006; Mellers, 2005), the diagnosis of NEAD has been observed to result in the immediate cessation of attacks (Farias, Thieman & Alsaadi, 2003; Reuber & Elger, 2003). Although it has recently been suggested there is no good quality evidence that receiving a diagnosis of NEAD has a significant (positive or negative) impact (Brough, Moghaddam, Gresswell & Dawson, 2015), further individual and group psychoeducation (e.g. Mayor et al., 2013; Zaroff, Myers, Barr, Luciano & Devinsky, 2004), were not reviewed, and have demonstrated reasonably positive outcomes in terms of attack reduction/cessation.

**Psychoanalytic/psychodynamic therapy.** Initially psychoanalytic case reports detailed NEAD treatment related to CSA (Goodwin, Simms & Bergman, 1979; Gross, 1979). More recently, an evaluation of brief (mean sessions= 6, range= 1-24) psychodynamic interpersonal therapy for patients with medically unexplained neurological symptoms (including NEAD) found 49.2% of patients significantly improved on at least one measure concerning either: emotional wellbeing, quality of life or somatic symptoms (Reuber, Burness, Howlett, Brazier and Grunewald, 2007). Long-term outcome of up to 20 sessions of psychodynamic therapy found that in 47 patients, 25.5% became attack free, a further 40.4% experienced a 50% or more reduction in attacks, and there was an overall reduction in health care utilisation (Mayor, Howlett, Grunewald & Reuber, 2010). Where appropriate however, CBT strategies were incorporated within the therapy in this study.

**Behavioural management and treatment.** Gardner (1967) reported on three sessions treating a child with NEAD. The therapy modified reinforcement contingencies operating within the family; providing the child with attention for appropriate behaviour but not for inappropriate behaviour including NEAs, resulting in rapid attack cessation. Though it would now be considered unethical, in a follow up when attention was reinstated for inappropriate behaviour attacks re-emerged, they subsequently ceased when attention was withdrawn.

Betts and Boden (1992b) reported on multi-modal treatment of 128 NEAD inpatients (including 46 with co-morbid epilepsy). Though most patients
received a combination of treatments, almost all were also treated with behaviour modification therapy; preventing rewarding of attacks by ignoring them, whilst deliberately rewarding other behaviour with verbal praise. Though outcome differences between treatments were not assessed, 63% of patients completed treatment attack free, 24% experienced a reduction, and the rest (13%) experienced no change or an increase. A two year follow-up found only 31% of those attack-free post-treatment had maintained this. As the behavioural treatment may have contributed to initial improvements, the authors suggested that environmental stress and rewards in the community may have contributed to losses at follow-up.

**Cognitive behavioural therapy (CBT).** Rusch, Morris, Allen and Lathrop (2001) reported on CBT treatment based on formulations of symptom patterns. For example, patients with an ‘anxiety/panic’ pattern received cognitive therapy with exposure, patients with a ‘post-traumatic stress and dissociation’ pattern received exposure therapy and patients with a ‘reinforced behaviour’ pattern received strategies involving family to modify reinforcement contingencies. 21/26 became attack free, and the other five had significant reductions in attack frequency. In a study of 20 patients, 12 CBT sessions resulted in attack reduction at a six month follow-up as well as improved social and occupational outcomes (Goldstein, Deale, Mitchell-O’Malley, Toone & Mellers, 2004). Following 12 sessions of manualised CBT, LaFrance et al. (2009) reported attack cessation in 11 out of 17 of treatment completers. Significant mean improvements were found in symptoms of depression, anxiety, and somatisation, and in quality of life and psychosocial functioning. In a pilot randomised controlled trial (RCT), 12 sessions of CBT in addition to standard medical care (SMC) was superior to SMC alone at treatment end (measuring attack frequency). Additionally, the CBT group was more likely to maintain attack freedom at a three month follow up (Goldstein et al., 2010).

A recent Cochrane review (Martlew, Pulman & Marson, 2014) systemically evaluated 12 studies with a total of 343 participants, and concluded there is little reliable evidence to support any therapy, including CBT, in the treatment of NEAD. However, it is important to consider that Cochrane reviews (used to develop healthcare policy) prioritise evidence according to amongst other
features statistical generalisability [see Case study research, pg 81], deeming RCTs and reviews of RCTs the gold standard research methodology. This means many other forms of evidence (held in higher regard within other policy areas; Davies, Nutley & Smith, 2000) are excluded, to the suggested detriment of advancing treatment and care (Roth & Fonagy, 1996; Slade & Priebe, 2001).

**Limitations of existing research.** The preponderance of psychodynamic interest in NEAD was suggested to be the reason for the delay in paying attention to other psychological processes involved in the development of NEAD (Francis & Baker, 1999). It has also been suggested that the focus on differential diagnosis has impeded the development of theoretical understanding and specific treatment approaches (Bodde et al., 2009; Brown et al., 2011). More generally, the discrepancy between NEAD burden and NEAD information (see Brigo & Igew, 2014) includes a lack of professional understanding (Shneker & Elliott, 2008), and difficulty agreeing where those with NEAD are best managed (Kanner, 2008), perhaps due to the preponderance of mind/body and mind/brain dualism amongst professionals (Miresco & Kirmayer, 2006; Salmon, 2007).

The validity of the identified risk factors can be brought into question when considering the limitations of the correlational research that proposed them (Bodde et al., 2009). A problem with reliance on cross-sectional designs, whereby NEAD and risk factors are measured simultaneously, is that it is difficult to determine whether these factors preceded or followed the onset of attacks. Such research has resulted in the identification of psychosocial factors more common in those with NEAD than in those with epilepsy as earlier detailed. However, the factors identified may be common across other clinical populations (Binzer, Stone & Sharpe, 2004; Bodde et al., 2011; Kabakoff et al., 1990; Krawetz et al., 2001; Rind, Tromovitch & Bauserman, 1998). Additionally, risk factors are relatively common in the general population (e.g. trauma: see Norris & Slone, 2013), yet NEAD is relatively rare (Benbadis & Hauser, 2000). The ubiquity of such factors calls into question their individual predictive validity and explanatory utility and raises the question of how they interact to produce NEAD.
The theories and models are predominantly descriptive incorporating the suggested risk factors, which as detailed appear to have limited explanatory utility. The mechanisms/processes by which suggested risk factors (or unidentified factors) interact in the development of NEAD remain unclear. Existing models have made efforts to explain development but either do not appear to offer a full explanation (acknowledging their shortcomings) (Baslet, 2011) or incorporate many concepts into a meta-model which is not specific to NEAD, and due to the proposition of numerous treatment targets is difficult to verify (Brown, 2004, 2006; Brown et al., 2011; Deary et al., 2007). The symptom modelling/learning theory and earlier psychoanalytic theories hold intuitive appeal but have not been subject to thorough application or verification and therefore remain unsupported.

It is therefore important that research attempts to identify any specific mechanisms underpinning NEAD in order to increase understanding of why it develops and persists, and later verify specific treatment approaches. Additionally, as NEAD is suggested to be a heterogeneous group, at least in presentation (Baslet et al., 2010; Gates et al., 1985), exploring the mechanisms through which the behaviour develops in specific cases may offer more information regarding the similarity or diversity of NEAD aetiology. The association between clinician’s lacking understanding, offering poor explanations to clients and poorer prognosis (Carton et al., 2003), also supports the need for further research into how non-epileptic attacks develop. Improved clinical understanding may facilitate clearer communication between professionals and patients.

There are clear general methodological issues with the evidence for NEAD treatment including mixed samples, poorly operationalised therapy and a distinct lack of control samples (Baslet, 2012; Bodde et al., 2009; Martlew, Pulman & Marson, 2014). However, a limitation specific to this population was highlighted by Reuber, Mitchell, Howlett & Elger (2005). They strongly advised against measuring attack frequency or cessation as the primary treatment outcome, as reductions in attacks are not always associated with other positive outcomes including occupational status and reductions in anxiety and depression.
Considering all of the literature and evidence it is unsurprising that previously employed nomothetic structural approaches, which seek to identify and describe features of phenomena, have failed to adequately conceptualise the complexity of NEAD. This indicates the need to explore whether a functional approach will offer more to understanding NEAD.

**Case study research.** Based on the reviewed literature and evidence, it appears that much of what is known so far is descriptive; What does NEAD look like? What experiences and traits have NEAD patients got in common? This information is no doubt useful, particularly as it has facilitated improved diagnostic accuracy; in the US correct diagnosis was associated with an average health care cost reduction of 84% within six months (Martin et al., 1998). What appears to be lacking are explanations, answers to ‘how’ and ‘why’ questions. For example, how does NEAD develop? and why does NEAD develop rather than any other symptom or behaviour? Yin (1994) suggests ‘how’ and ‘why’ questions are best addressed through case study research.

Bromley (1990) describes a case study as “a systematic inquiry into an event or a set of related events which aims to describe and explain the phenomenon of interest” (p. 302). Despite criticism of case study research, that at best it provides interesting presentations of unique cases, Bromley (1986) proposes that being sensitive to uniqueness is a strength of case studies over cohort studies. By analysing cases individually researchers are able to modify initial conceptual frameworks in response to convergent and divergent features arising in new cases (Bromley, 1990).

Typically research which proffers statistical generalisation, that is attempting to apply results from a particular sample to a whole population, has been considered most useful (Firestone, 1993). Case studies have been misunderstood and critiqued, among other reasons, due to their inability to offer this type of generalisation (Flyvbjerg, 2006). Case studies are however, able to offer analytic/theoretical generalisation, considering in-depth findings in relation to theoretical propositions (Verschuren, 2003). Many case studies, however, are structural in orientation, describing the presence and/or absence of events and characteristics surrounding or comprising a particular problem/disorder.
Case studies which seek to move beyond description to explain the development of a problem adopt an approach to formulate/conceptualise the information. Many approaches (e.g. cognitive behavioural) rely on introspection and infer the existence of unobservable structures making reliable measurement and scientific inquiry difficult to apply (Sturmey, 2008). Such approaches also minimise individual differences, for example assuming that internal thoughts are common across those with a similar presenting problem. This may be a misguided approach to understanding psychopathology, particularly when its development and characteristics appear to be heterogeneous (Dougher & Hayes, 1996) as has been suggested with NEAD (Baslet et al., 2010). An approach that is explanatory, which does not rely on introspection, and takes full account of individual differences is functional analysis (Sturmey, 1996).

**Behavioural case formulation: functional analysis.** Functional analysis (FA) is an approach to case formulation that allows contemporary phenomenon to be analysed in the context of an individual’s environmental and learning history (Sturmey, 1996). Functional analysis is based on the discipline of applied behavioural psychology and the principles of classical and operant conditioning (Pavlov, 1941; Skinner, 1953, 1974) [A glossary of terminology can be found in the Journal Paper Table 5. pg 38, and here Table 11. pg 95]. Early behaviourism was concerned with the analysis of observable behaviours only, a position which drew criticism from researchers concerned with mentalism (see Chomsky, 1959). However, Skinner’s (1953) approach to human behaviour, termed ‘radical behaviourism’, encouraged the analysis of cognitive experiences in the context of observable behavioural contingencies. Cognitive and affective experiences (covert behaviours) are distinguished from observable behaviours (overt behaviours) in this model, and both are developed and maintained by an individual’s interaction with the environment. Skinner proposed that over time a repertoire of learnt covert and overt behaviours are developed based on interaction with and reinforcement from the environment. This constitutes an individual’s learning history (Skinner, 1974). An individual’s learning history and behavioural repertoire can be understood by means of a functional analysis (Sturmey, 2008). The main benefit of using functional analysis is to develop an
idiosyncratic understanding of how an individual’s behaviour(s) have developed over time.

Functional analytic case formulation has been suggested as a research tool (Sim, Gwee & Bateman, 2005) and the use of functional analysis within single case designs previously supported the development of operant conditioning research (Morgan & Morgan, 2001) and aggregation of single participant research to generate hypotheses about causality is used in other disciplines such as medicine (Nuland, 1988). Functional analysis has been used to advance understanding of a wide range of complex psychological phenomena (Hanley, Iwata, & McCord, 2003), including: depression (Kanter, Cautilli, Busch, & Baruch, 2005), domestic violence (Bonem, Stanely-Klime, & Corbin, 2008), eating disorders (Slade, 1982), recidivistic arson (Jackson, Hope, & Glass, 1987) and self-injury (Bachman, 1972; Iwata, Dorsey, Slifer, Bauman, & Richman, 1994; Nock & Prinstein, 2004).

**Study aims.** From a methodological stand-point, the research aimed to address the limitations of nomothetic, structural approaches (predominantly cross-sectional cohort correlation studies) by applying an idiographic functional approach to understanding how and why NEAD develops.

This study aimed to use the case study methodology Multiple Sequential Functional Analysis (MSFA) to examine the development of non-epileptic attacks in the individual life trajectories of a small group of adults with NEAD. Furthermore, it aimed to compare and contrast these trajectories to generate hypotheses about the potential functions of non-epileptic attacks for these individuals, which may contribute to future research regarding theory and treatment.

**Methodology**

**Research questions.** The questions guiding the research were:

- How do non-epileptic attacks appear to develop in the histories of a sample of adults diagnosed with NEAD?
- What are the functions of non-epileptic attacks for these individuals?
• How do previously suggested risk factors appear to interact to influence the development of NEAD in these individuals?
• Are there similar pathways in the development of NEAD for the different individuals?
• Do the non-epileptic attacks have similar functional qualities for the different individuals?

**Ethical approval.** This study was granted ethical approval by the WALES/4 NHS research ethics committee, the research and development departments of three participating NHS trusts, and the School of Psychology Research Ethics Committee (SOPREC) at the University of Lincoln [documents attached in Appendix A]. Ethical considerations can be found in the ethical application to SOPREC [Appendix A] and the study protocol [Appendix B].

**Epistemology.** Epistemology is important to consider as it is the basis of how knowledge development is approached (Anastas, 2002). The epistemological position of a researcher will affect their research design, conduct, and how they interpret the results (Potter, 1996).

The epistemology underpinning this study is functional contextualism (Biglan & Hayes, 1996; Gifford & Hayes, 1999; Hayes, 1993; Hayes, Strosahl & Wilson, 1999). The contextualist philosophical worldview is that events are interpreted as ongoing acts inseparable from their context, this root metaphor is known as ‘act in context’ (Fox, 2008). Within this, functional contextualism’s primary goal is to predict and influence events with precision, scope, and depth in order to construct practical knowledge (Biglan & Hayes, 1996; Fox, 2006, 2008). The pragmatic truth criterion considers truth in relation to effective action, an analysis is considered true to such an extent it leads to effective action, or achievement of a goal (Fox, 2006; 2008).

Functional contextualism underlies modern behavioural psychology embracing the scientific principles of learning and conditioning (Fox, 2006). Indeed, Skinner exemplified the pragmatic truth criterion describing scientific knowledge as rules for effective action, in which truth can be considered if rules yield the most effective action (Skinner, 1974). Complimentary to theory and subsequent
treatment development, functional contextualists study behaviour in its current and historical context in an effort to construct principles and rules that are able to predict and change that behaviour in a variety of settings (Fox, 2008). Though functional contextualists favour experimental research which can manipulate variables to measure their influence on an event/phenomenon, any methodology that contributes to pragmatic goals is valued (Biglan & Hayes, 1996; Hayes, 1993). The contextualist root metaphor suggests that although current context of behaviour/action is important, to understand the purpose, meaning and function, historical context must be particularly appreciated (Morris, 1997). This fits with the MSFA methodology as it involves studying the life histories of participants in order to understand current behaviour and how it has developed within changing contexts.

Functional contextualists limit the context of events to be studied, which could be potentially infinite, based on the contextual features which aid achievement of (are pragmatic to) the goal (Fox, 2008). The purpose/goal of this analysis was to predict the influence of events and psychosocial factors to produce behavioural explanations of NEAD development. In line with behavioural principles of learning this focused the data collection on context preceding onset, and preceding and following attacks in the participants’ histories. Though the suggested psychosocial factors in the literature appear to be common place and non-specific, they may interact in the development of NEAD and therefore if/when they were reported by participants they were a focus of analysis. Analysing the development in each adult individually allowed consideration of different contexts in which NEAD can develop. The study also considered the function of participant behaviour (including verbal and non-verbal behaviour within interviews) within its context, and aimed to make links between the behaviour and all available data, to achieve a coherent working understanding of the development and maintenance of non-epileptic attack behaviour for each participant.

**Study design.** Functional analysis is a behavioural method which attempts to understand the function of behaviour by identifying variables which strengthen or reduce the likelihood of a specific behaviour occurring. A particular behaviour (or ‘target behaviour’) is understood through the use of an
**A:B:C:** analysis. ‘A:’ is the antecedent or triggering environmental event, ‘B:’ is the covert and overt behavioural response and ‘C:’ is the environmental consequence of the behaviour (Sturmey, 1996; 2008). A chain of A:B:C: analyses can be used to give a dynamic understanding of an individual’s learning history, where one analysis becomes the antecedent of the next sequence (Gresswell & Hollin, 1992). Identifying a functional relationship between variables in this way does not assume causality, but the order of events is both necessary and sufficient, known as temporal precedence, to assume that a functional relationship exists (Haynes & O’Brien, 1990).

Experimental functional analysis which could test the predictions made by the MSFAs and establish causal factors/mechanisms of NEAD development, was outside the time limits of this thesis project. However, retrospective biographical interviewing can gather in-depth data required for a descriptive functional analysis (Anderson, 1981; Sturmey 1996). Rather than being biased by the specific suggested ‘risk’ factors, biographical interviewing encourages all potentially relevant information to be elicited. In addition to reducing the impact of assumptions based on prior knowledge, it also gives more opportunity to identify unique experiences and factors (Krauss, 2012). A descriptive functional analysis leads to the generation of hypotheses to be verified by future, more focused research (Sturmey, 1996). With little known about the mechanisms and functional development of NEAD, this was deemed an appropriate and potentially useful method at this time.

A form of functional analysis that has been used as a case study research methodology is MSFA (Gresswell & Hollin, 1992). MSFA applies an established behavioural model, operant learning (Skinner, 1974), to provide a framework for understanding the functional development of behaviour across the life of an individual. Using case material from multiples sources, a chain of A:B:C: functional analyses are developed, linked by the proposition of key learning experiences which are hypothesised to have influenced the development of the target behaviour across time. This sequential analysis generates explicit hypotheses about the functional relationships between events and behaviour (Gresswell & Dawson, 2010; Gresswell & Hollin, 1992), and has been successfully used to facilitate understanding of the development of complex
behaviour, including: multiple murder (Gresswell & Hollin, 1992), violent behaviour (Hart, Gresswell, & Braham, 2011), offence paralleling behaviour (Gresswell & Dawson, 2010) and female perpetrated intimate partner violence (Mappin, Dawson, Gresswell & Beckley, 2013).

MSFA is an intensive methodology collecting a comprehensive amount of data from multiple sources. In line with other MSFA studies (Hart et al., 2011; Mappin et al., 2013), between three and six participants were sought. Due to the intensive nature of the method, it was believed that this would be sufficient to capture a potential range of learning sequences/pathways to the development of NEAD.

The gap in the literature as earlier detailed is a comprehensive understanding of the mechanisms by which NEAD develops across the life span. Whilst qualitative methods are useful for establishing themes/patterns, they would not enable the identification of psychological mechanisms as MSFA is able to. The use of MSFA to identify the psychological mechanisms of NEAD aimed to improve the understanding of the disorder which may in turn (by informing future research) improve assessment and treatment. Qualitative methods, whilst potentially useful in identifying ‘what’ (what themes/experiences are related to NEAD), are not able to suggest ‘how’ (how do these themes/experiences influence or lead to the development of NEAD). With the limitations of both quantitative and qualitative methods in understanding NEAD explained, MSFA was the pragmatic choice for this research.

Bromley (1986) described criteria which must be met for case study research to be considered a worthwhile scientific enterprise:

1. It must give an **explanatory account** of the reasons for behaviour. The research aimed to produce an explanatory account of the development and maintenance of NEAD, underpinned by the behavioural principles of operant learning (Skinner, 1974).

2. It must aim to **improve knowledge** by providing new information which can be drawn on by future researchers. The research aimed to add to existing knowledge by using a method novel to exploring
the development of NEAD which may identify important new information to be examined in future research.

3. It must **develop or sustain the discipline** of studying individual cases. Applying MSFA to understanding NEAD develops the discipline of studying individual cases by adding to the assessment of the utility of this research method.

4. Depend on **acceptable procedures** and arrangements.
The procedure for MSFA is well-established and the more general research procedure and conduct was considered through university and NHS boards of ethics, and research supervision.

**Participants.** Three participants were recruited. All participants and their relatives/friends were given pseudonyms for the purposes of this study, other details have been generalised to protect the participants’ identities [A summary of demographic variables are presented in the Journal Paper, Table 4 pg 36]. Further to this, a brief contextual summary for each participant is offered below.

Jayden, the only male recruited was 30 years old and was in a seven year relationship with his partner with whom he had a three year old daughter. Susan was a 62 year old married mother of four and grandmother. Daisy was a 31 year old married mother of two daughters.

In addition to a diagnosis of NEAD, Jayden also had current diagnoses of medication overuse headaches and depression. Susan also had diagnoses of functional neurological disorder (paralysis), chronic pain, and fibromyalgia. Daisy had additional diagnoses of myalgic encephalopathy (chronic fatigue syndrome) and fibromyalgia.

**Recruitment.** A Consultant Neurologist working across two NHS trusts was asked to identify outpatients with a diagnosis of NEAD. Those who attended an outpatient clinic appointment with him and met the study criteria were asked if they would be interested in taking part, and were given the participation information sheet [see Appendix C].
The information sheet asked interested people to contact me by telephone or email. The three participants who expressed an interest by contacting me were eligible to participate and completed the project.

Acknowledging the substantial time commitment of the lengthy and multiple interviews, the study offered financial reimbursement for participants. Participants were offered a £5 gift voucher of their choice for every hour of their contact time with me. This was not dependent on completion and had anyone withdrawn they would have received their vouchers to the value of their participation to date.

A consecutive recruitment strategy was employed, continuing until either the maximum number of participants was reached, or the timescale dictated there was too little time to complete the process with another participant (and at least three participants’ data has been gathered to the point where they were unable to withdraw it). Three people were recruited when the timescale suggested there would be too little time to complete the process with any more participants. The three people interested in the study participated; no additional prospective participants had to be turned down.

**Inclusion criteria.** Identified prospective participants were eligible for participation if they were 18 or over with a diagnosis of NEAD and were attending outpatient neurology services in the identified NHS trusts. Relatives/professionals also had to be 18 or over.

**Exclusion criteria.** Participants and relatives/professionals would be excluded if they were unable to communicate and understand English spoken language as assessed by the Consultant Neurologist. This was due to the in-depth nature of the interviews which comprised the majority of the study data. The constraints of the study budget did not allow for the expense of a translator/interpreter. Additionally, considering the non-verbal behaviour of participants, a triadic relationship in the interviews may affect the potential data or analysis.

Participants who would not consent to their files being accessed would be excluded from the study due to the reliance on triangulation in the analysis. For
the same reason, participants who could not identify, or would not consent to, a relative/professional being interviewed, would also be excluded from the study.

Consent. During an initial telephone call the researcher answered any questions prospective participants had. An initial meeting was arranged (at an NHS site) where further information about the study was shared and any questions answered. Prospective participants were informed that expressing and interest and attending the meeting did not constitute an agreement or obligation to take part. Confidentiality limits, that any disclosure or concern about current risk to the participant’s or other’s safety may have to be reported, were explained. The potential for the interviews to evoke strong emotion and distress was discussed and a plan for each participant to access support through their current care network or new referral, if required, was agreed. Prior to the initial meeting I knew only the name, contact details, and hometown of each participant, and that each had received a diagnosis of NEAD from the recruiting Consultant Neurologist.

At this point the prospective participants were asked to give informed consent to take part in the individual, audio recorded interviews, to allow the researcher to conduct a file review of relevant documents and records, and to identify someone who knows them well to be interviewed with regard to their history and NEAD [see Appendix C].
**Procedure.**

**Figure 2.** The data collection and analysis procedure for each case.

**Interview procedure.** Participants were interviewed in one-to-one sessions, each lasting around 90 minutes, for between five and seven hours. The initial interview was conducted at an NHS site and subsequent interviews were either at an NHS site or in the participant’s home. Plans were discussed with the participants in the event of them having an attack during the interviews.
The first two interviews gathered a detailed developmental history from the client. Following this an initial MSFA was developed. The latter two-three interviews focused on gathering information to fill gaps in the history and more detail regarding the areas already deemed relevant to the developing MSFA. The interviews loosely followed the developed semi-structured interview guide [see Appendix D], outlined at the request of the University of Lincoln School of Psychology Research Ethics Committee (SOPREC). The broad areas covered included childhood, relationships, school, work, and illness/injury. Each interview ended when sufficient information was gathered relevant to that stage of the process. Additionally, due to participant’s physical symptoms, their fatigue was also considered with session length responsive to their needs.

All interviews were audio-recorded and I took anonymised notes throughout, all data was stored in accordance with University policy. The interviews did not need to be transcribed as no textual analysis was to be undertaken.

**Relative interview procedure.** Each participant and I discussed the remit of the supplementary interview and identified a person who knew them well. Participants were given the additional information sheet to pass onto the identified relative [see Appendix C]. The aims of the study and the specific interview, and confidentiality limits were explained in more detail in person. All those initially identified were happy to take part and provided informed consent [see Appendix C]. Jayden’s mother, Susan’s best friend, and Daisy’s husband were interviewed. A broad guide for these interviews is included in Appendix D. The focus depended on the developed MSFA as these interviews aimed to verify and triangulate information already provided by the participant. These interviews were also audio-recorded, notes were made, and they lasted between one and two hours.

**File review.** Files reviews were completed for each participant, which allowed existing information to be triangulated and also allowed the MSFA to be checked against the hypotheses and suggestions of other professionals (consensus checking). The documents reviewed for each participant are presented in Table 10.
Table 10. File documents reviewed

<table>
<thead>
<tr>
<th>Jayden</th>
<th>Susan</th>
<th>Daisy</th>
</tr>
</thead>
<tbody>
<tr>
<td>Psychology clinical notes</td>
<td>A&amp;E documents</td>
<td>Stroke clinic documents</td>
</tr>
<tr>
<td>Psychiatry outpatient clinic</td>
<td>Orthopaedic clinic</td>
<td>Psychiatry report</td>
</tr>
<tr>
<td>letters</td>
<td>letters</td>
<td></td>
</tr>
<tr>
<td>Neurology clinic letters</td>
<td>Neurology clinic letters</td>
<td>Neurology clinic letters</td>
</tr>
</tbody>
</table>

**Final session.** Each participant attended a final meeting, where the chronological order of their life events relevant to the development of NEAD was shared. Participants were asked to give feedback on the accuracy of the data gathered and the order of events.

**Reflection.** I kept a reflective diary after each interview/data collection session and after supervision when relevant. This was used to facilitate reflexivity and transparency in the process by recording my thoughts, assumptions, and subjectivities which may have influenced the process. The reflective diary was also analysed using MSFA in order to consider how my learning experiences through the process may have influenced my subsequent behaviour [see The experience of doing the research (my MSFA), pg 143].

**Triangulation.** Qualitative methods typically rely on data from individual introspective interviews, which can be considered to limit the scientific validity and reliability of the results (Silverman, 1989). Additionally, research relying solely on self-reports, particularly concerning emotional experiences, have been criticised due to diminished insight and awareness of internal states in those with alexithymia and those who avoid (found in significantly more NEAD patients than in epilepsy and control groups; Di Maro et al., 2014).

Triangulation is supported in case study research (Yin, 1984) and is used in MSFA to gather data relevant to complex behaviour which has developed over time (Gresswell & Hollin, 1992). Triangulation in this study aimed to synthesise data from multiple sources to form a comprehensive narrative and explanation of the development and maintenance of NEAD for each participant.
Data and method triangulation were utilised; gathering verbal and written information from different sources regarding the same phenomenon (Sayer, 1992). In this study the data sources were interviews with participants, relative interviews and file reviews. The behavioural theory underpinning functional analysis also acted as source for triangulation, which was particularly useful in resolving discrepancies within and between the other sources of data. This was achieved by considering the context and information either side of the discrepancy chronologically. This enabled predictions to be made regarding the likely influence of each option on known future events/behaviour and therefore reach a decision. To reduce the potential researcher bias discrepancies were discussed in detail with the research supervisors.

**The behavioural processes and language underpinning MSFA.**
Radical behaviourism and the science of applied behavioural analysis (Skinner, 1953, 1974), from which MSFA was developed, propose rules about the development of behaviour. These rules are borne out of comprehensive empirical studies of both animals and humans. Particular terminology is used to convey these rules and concepts.

As the behavioural knowledge of the journal readership could not be assumed, it was deemed necessary to provide an overview of the terminology applied in the current study [see the Journal Paper, Table 5, pg 38]. Additional terminology used within the extended results and analysis are defined in Table 11.

For ease of understanding the main presentation of the analyses are jargon-free narrative descriptions of the participant’s lives (Tables 12 to 14). Within the descriptive narratives behavioural terms are noted and following each narrative these are presented within behavioural explanations and hypotheses incorporating relevant NEAD literature.
Table 11. Additional glossary of behavioural terms

<table>
<thead>
<tr>
<th><strong>Discriminative stimulus</strong></th>
<th>Antecedents that must be present for the specific behaviour to be emitted</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Establishing operation</strong></td>
<td>A setting event/context that increases or decreases the value of a reinforcer e.g. hunger</td>
</tr>
<tr>
<td><strong>Continuous reinforcement</strong></td>
<td>Reinforcement occurs following every occurrence of the behaviour. A rewarding stimulus may become less effective at reinforcing behaviour as it becomes less appealing e.g. a chocolate bar becomes less appealing after the tenth one and therefore the behaviour of eating the next one is less likely.</td>
</tr>
<tr>
<td><strong>Intermittent reinforcement</strong></td>
<td>Reinforcement for behaviour vary by time interval or frequency. Patterns can be fixed or variable in terms of the time interval or behaviour frequency ratio. Variable schedules tend to be most resistant to extinction* due to the unpredictable occurrence of reinforcement.</td>
</tr>
<tr>
<td>*<strong>Extinction</strong></td>
<td>The eventual cessation of a behaviour due to the discontinuation of reinforcement.</td>
</tr>
</tbody>
</table>

Though, covert and overt behaviours are presented separately, this was a pragmatic decision made to facilitate clearer presentation and parsimony in the analyses. It does not imply causality/directionality or that cognitive events are separate to or precede observed events in terms of experience. Whilst from a behavioural theoretical stand-point emotion is considered to be a complex combination of physiological experience, behaviour, context, and cognition (Skinner 1974), for similar reasons feelings/emotions are presented using commonly understood descriptive labels e.g. anger.

**Analysis.** MSFA (Gresswell & Hollin, 1992), embedded in the methods, evidence base, and philosophical assumptions of radical behaviourism, was the
analysis used in this study [the Journal Paper details the analysis procedure in more detail in order to offer transparency and replicability of the process, pg 37]. From recruitment through to the completion of the final case conceptualisation (comprised of the MSFAs) took six months.

Developed by Gresswell and Hollin (1992), MSFA builds on functional analysis by organising information into a series of A:B:C:s to account for learning through complex chains of behaviour. It represents a developmental process whereby one A:B:C: explicitly influences the (A:) antecedents of the next, aiming to demonstrate the influence of previous events on subsequent behaviour. In line with radical behaviourist principles, behaviour (B:) includes that which is overt (directly observable) and covert (thoughts, feelings, and physiology). As with functional analysis, MSFA does not purport to make statements of causality, however, the ordering of complex material can lead to explicit hypotheses based on the temporal relationships between variables (Haynes & O’Brien, 1990). A summary of learning (key learning) as a result of each developed A:B:C: is hypothesised, to explain how the participant’s repertoire of behaviour may have changed as a result of the specific learning experience. The process was considered complete when the research supervisors and I determined that the life history and resulting MSFA produced a theoretically coherent and complete understanding of the development of the participant’s non-epileptic attacks.

Qualitative research methods have been criticised for merely producing a list of themes; Ayres, Kavanaugh, and Knafl (2003) advise that stand-alone themes have no explanatory power, without demonstrating how they work together data analysis is incomplete. The MSFA process constitutes a within-case analysis as it hypothesises the relationship between factors/events which have led to the development of non-epileptic attacks. Considering the suggested heterogeneity of the NEAD population; an across-case analysis may offer new understanding of different/similar pathways and factors in NEAD development. Once the MSFAs were complete they were reviewed for similarities and differences in: historical factors, reinforcement schedules, and functions. Utilising within- and across-case analysis, this research aimed to enable the presentation of individual cases in a potentially generalisable way.
Results and discussion of analysis

The results and discussion section is comprised of the MSFA sequence for each participant (Tables 12, 13 and 14). Each is followed by an analysis which is intended to explain the key behavioural principles in developing the MSFA.

Jayden. Jayden is a 30 year old, white male who has been experiencing non-epileptic attacks since the age of 11. Epilepsy was diagnosed aged 11 and a revised NEAD diagnosis was made six years ago. He has been with his partner for eight years and together they have a four year old daughter. Jayden lives with his mother and step-father due to the perceived unpredictability of his attacks and related aggressive behaviour. Jayden was recruited as he attends a neurology outpatient clinic in relation to his NEAD.

Table 12. MSFA for Jayden

Early experiences

Jayden, is the youngest of four siblings (two sisters, one brother), and was brought up in an upper working class family with his mother and father. His father worked in the military. His mother worked part-time and also took on the traditional housewife role. Jayden described his community as one where the men gave the women money for housekeeping and spent the rest at the pub, where they spent a lot of their time.

Jayden described being treated differently to his siblings by his father who he reports was physically violent towards him from as early as he can remember. Being in control of the money his father also ensured that he didn’t get the same gifts and material possessions as his siblings. Jayden reported that the violence was always when his father was drunk, and feels it was because his father suspected that he was not his biological child. Jayden also suspected this recalling memories of his mother having affairs from an early age. When he was around eight years old Jayden found out that his father had had other children before meeting his mother.

Jayden felt angry and resentful towards his father for the way he was treated. He reported coping by suppressing his anger, and becoming avoidant and isolated within the family home. The family did not react to this withdrawal which Jayden said suited him but now he reflects that because of this his childhood was generally “horrible”. His parent’s relationship was volatile with violence on both parts, which Jayden witnessed from an early age (usually instigated by his mother when his father came home drunk having spent too much money). Jayden described his community as rural with very few other children his age around and little to do. His activities
were very solitary; his siblings had different interests due to their ages (the youngest being three years older than him and the oldest eleven years older).

Jayden dealt with his father's violence by submitting and then when he was able to (if his father was too drunk and exhausted) by running away. His father was never violent to him in his mother’s presence, though she was often at work. Jayden reported that he lied to his mother about injuries his father inflicted on him if they were noticed or obvious. Jayden feared that if he told the truth it would cause more conflict between his parents which was bad enough already. Jayden described his mother as being caring, particularly if he was poorly. One of his earliest memories is him being poorly and being allowed to curl up on his mother’s lap and cuddle her.

**Functional analysis sequence 1**

**Antecedents**
Jayden is singled out by his father, receiving less attention and possessions than his siblings, and being the only one subjected to drunken physical abuse on a regular basis. Jayden's mother was unaware of the abuse.

**Behaviours**

**Covert**
No one can protect me (depression)
I am unimportant, no one notices me or what’s happening to me (sadness/apathy)
It is unfair that I am being singled out, others are treated better than me (anger/resentment)

**Overt**
Submit or flight response to violence (avoidance)
Withdrawal in the home
Some comfort seeking behaviours - reporting illness to mother

**Consequences**
No positive social relationships are developed
Emotional needs not met
Positive interaction/attention with mother when physically unwell and father isn't present (approach/avoidance conflict with illness behaviour positively reinforced)
Illness serves to reduce risk of being beaten as mother is then present (illness behaviour negatively reinforced)

**Key learning**
- Life is unfair, (being singled out/treated worse than siblings is unfair).
- Being unwell leads to being comforted.

________________________________________
Early adolescence and school
With no previous learning of positive social relationships Jayden reported finding it difficult to engage with peers at school. When he was verbally bullied seemingly as he was judged an outsider he would easily react, resulting in physical fights and a reputation not to mess with him. Jayden missed school often due to migraines which began when he was around nine, after his father threw him down the stairs (though he reported that he fell). If a migraine started at school Jayden would either walk home, or if his migraine was really bad he would ring his Mum who would leave work to pick him up and take him home. When at school Jayden reported that he was bored by the easy work (reportedly being particularly good at maths), and was regarded ‘a smart arse’ by his teachers as he would ‘back-chat’ them. Jayden reported that he was happier in his own company and that he would go to bed when he experienced migraines. Jayden’s father continued to drink and physically assault him. At home Jayden also began to “act out”, he would swear and refuse to do things his mother asked him to. Jayden reported that his mother was strict on bad behaviour and would smack him in such situations.

Functional analysis sequence 2
Antecedents
Key learning from sequence 1
Father’s violence continues, including throwing Jayden down the stairs
Social demands on Jayden increase in the school environment
Jayden is verbally bullied

Behaviours
Covert
I am unfairly treated (anger strengthened)
I don’t get on with others I am better off alone
Overt
Reacts to verbally bullying with physical violence
Expressing anger through “acting out”: swearing and refusing to do what is asked of him.
Migraines
Phone Mum to elicit care
Goes home to bed (withdrawal continues)

Consequences
Jayden earns a reputation for physical fights and bullies retreat (aggression positively reinforced).
Jayden’s “acting out” is positively punished at home, being hit by Mum.
Illness behaviour is positively reinforced by receiving increased care from Mum.
Jayden’s illness behaviour and subsequent withdrawal is negatively reinforced – avoiding negative interactions at school and home.
Key Learning

- Life is unfair, (result of being targeted by peers and father).
- Aggression is respected and it makes people stay away
- Avoiding others makes life easier.

Head injury and seizures
Jayden began playing rugby at school in year seven – he was physically well-built and his aggressive attitude had been recognised by the coach. He was encouraged to use these to his advantage in rugby, he enjoyed it and received praise for his abilities. His social interactions remained limited and he continued to have regular absences from school due to migraines. Aged 11 during a rugby match Jayden was brought on to “take someone out” as the other team was winning. Jayden aggressively tackled a player and in turn he was piled on by five players. Jayden was reportedly knocked unconscious and suffered a seizure characterised by jerking and “foaming at the mouth”, reports suggested that he was "out cold" for around 10 minutes. An ambulance was called and he was taken to hospital. Following this Jayden had two weeks off school and spent time with his mother, during this time his father did not physically assault him. Jayden was in bed a lot, feeling dizzy and sick and had nearly daily unpredictable seizures during this period.

Upon returning to school Jayden continued to play rugby despite medical advice not to. When he experienced seizures during rugby the school refused to allow him to play. As the seizures continued investigations were conducted, including computerized tomographic (CT) scanning, resulting in a diagnosis of epilepsy around six months later. Jayden was then put on anti-epileptic medication.

Functional analysis sequence 3
Antecedents
Key learning from sequence 2
Jayden is praised by his rugby coach and respected on the pitch for his aggression
Jayden is knocked unconscious and experiences a seizure
Jayden is signed off sick from school to rest
Jayden is diagnosed with epilepsy

Behaviours
Covert
I am good at something and people recognise it (pride)
It’s not fair that I can’t do what I want (frustration)

Overt
Post-head injury seizures continue
Consequences
Seizures can lead to injuries and embarrassment due to incontinence (aversive)
Jayden avoids negative social interaction by being at home off sick (seizure behaviour negatively reinforced)
Positive interaction with mother increases (seizure behaviour positively reinforced)
Being around mother means physical violence from father reduces (seizure behaviour negatively reinforced)
Positive interaction through rugby is removed (seizure behaviour is negatively punished)

Key Learning
• Expressing anger can be unsafe (due head injury and resulting seizures)
• Overall seizures reduce negative experiences and increase positive experiences.
• Sense of worth and value derived from praise for rugby and increased care from mother.

Fight with Father
When Jayden was nearly 12 he was on anti-epileptic medication, his seizures had reduced in frequency. He was back at school (around 2 days per week due to continued migraines and seizures), though he was still not allowed to play rugby. Based on the reported events, frequencies and semiology, it is hypothesised that Jayden experiences both epileptic seizures and non-epileptic attacks [see Journal Paper section 4.4 for more information, pg 43]. It is hypothesised that by the time the epileptic seizures were better controlled the behaviour had been conditioned by its consequences and non-epileptic attacks began.

Though Jayden would usually ask his mother for dinner money one morning he asked his father (his mother was in bed after working a night shift). His father was still drunk from the night before, he didn’t know Jayden had been receiving dinner money as he was supposed to be utilising free school meals, and he hit out at Jayden. Jayden reported feeling an overwhelming rage, and perhaps influenced by increasing emotional lability/decreased inhibition (following his head injury), and an increased sense of self-worth, he fought back against his father punching him. His father appeared to be in pain and it became apparent that he was having a heart attack, which killed him.

Functional analysis sequence 4
Antecedents
Key learning from sequence 3
Seizures are better controlled by medication
Father is physically violent towards Jayden again
**Behaviours**

**Covert**

This is not fair (injustice, rage)
I am not standing for this anymore (sense of worth resulting from praise for rugby and increased care from mother)

**Overt**

Non-epileptic attacks begin
Jayden expresses anger by punching his father

**Consequences**

Non-epileptic attacks are reinforced in the same way seizures are (care from mother, and resultant protection from father and avoidance of school)
Father has heart attack and dies

**Key Learning**

- Expressing anger is unsafe (consolidation)
- Suppressing emotions is best
- Having seizures (and now non-epileptic attacks) enables avoidance which means anger is less likely.

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**Non-epileptic attacks and continued restrictions on life**

Following his father's death Jayden explained to his mother that his father had hit him and the truth was revealed about the years of physical abuse he had been subjected to. Jayden’s mother felt extremely guilty about this and tried to make it up to Jayden by spoiling him and allowing him to “get away with anything” which Jayden reported continues to this day. In the weeks following his father’s death Jayden’s non-epileptic attacks increased in frequency significantly (from fortnightly to daily). Jayden reported that his non-epileptic attacks are characterised by staggering and falling, and thrashing movements (which can cause damage to property or injury to people) and disorientation coming out of it (Jayden has lashed out if he is unsure where he is and who he is with). Jayden’s life continued with years of contact with medical professionals due to his continued attacks/seizures. His life was restricted by his epilepsy diagnosis, he was not able to learn to drive and begin a career like his peers and siblings. He worked for short periods but was let go/sacked due to his attacks or high levels of sickness. He continued to experience migraines which added to his frustrations. The side effects of medication, including weight gain and hair loss, also frustrated him. Jayden’s anger about the impact of medication and restrictions of the diagnosis increased as he was seen by many medical professionals and the medication wasn’t helping, as non-epileptic attacks occurred regularly. Jayden became so frustrated, he threatened to punch his Neurologist when
it was suggested that Jayden’s attacks were “all in his head”. As a result he was discharged from the service and he was unable to get his prescribed medications, and he feared things would get worse without medication. Jayden reported that his mother made a complaint about this and he was subsequently referred to see a different neurologist.

**Functional analysis sequence 5**

**Antecedents**
Key learning from sequence 4
Not able to do things peers are doing
Physical side effects and symptoms
Being told what to do/not to do by professionals

**Behaviours**

**Covert**
It's not fair (anger and frustration)
I'm not getting better (anger)
Feeling like this is intolerable (anger/depression)

**Overt**
Non-epileptic attacks increase in frequency
During attacks feeling disorientated Jayden hits people who are trying to rouse him
During attacks Jayden damages property when falling or jerking/thrashing
Jayden tells his mother the truth about the physical abuse
Jayden threatens to punch neurologist (expressing anger)

**Consequences**
Non-epileptic attacks do not lead to incontinence (less embarrassing therefore less aversive)
Non-epileptic attacks are reinforced as before but additionally result in avoidance of the feared consequences of expressing anger
Friends and relatives become fearful around Jayden
Non-epileptic attacks and reporting the abuse increases care from mother
Jayden is discharged from the Neurology service (Jayden fears things will get worse)
Jayden’s mother complains and he is given a new Neurologist and his medication continues

**Key Learning**
- Expressing anger is unsafe
- Being around others isn’t safe.
- Avoiding people and suppressing emotions is best for everyone

**Current context**
Jayden began a relationship with his current partner when he was around 22 years old. When
Jayden was around 24 years old he was referred to his current Consultant Neurologist who assessed his attacks and diagnosed NEAD. Jayden felt listened to describing him as the first professional to listen to him and not patronise him. As a result of this revised diagnosis, Jayden’s medications were changed and over time there have been slight reductions in his migraines and seizures. Jayden and his partner had a child together three years ago. Jayden reported finding maintaining his relationship challenging. He reported that his partner’s need to be close to him clashes with his need to be alone. At times he has felt as though he isn’t able to give her everything she wants but he gets angry when she pressures him to do more than he feels he is able to. Jayden also described being fearful of having an attack in front of his daughter in case she gets hurt. Because of this Jayden will not live with his partner and child, instead he lives with his mother and step-father. Living separately is reported to exacerbate his partner’s neediness which he finds difficult to tolerate.

**Functional analysis sequence 6**

**Antecedents**
- Key learning from sequence 5
- Jayden lives with his mother and step-father
- Jayden’s relationship is strained
- Jayden’s partner makes requests for him to spend more time with her and their child

**Behaviours**

**Covert**
- She knows about my difficulties, she should not be asking me to do this (anger and frustration)
- It’s not safe for me to be around people when I am feeling like this (fear)

**Overt**
- Withdraw when possible (to mothers house)
- If withdrawal not possible a non-epileptic attack may occur

**Consequences**
- If Jayden withdraws, demand soon continues and anger grows
- If Jayden has an non-epileptic attack he is left alone for longer to recover (attacks are negatively reinforced, more so than withdrawal, by reducing anger)

**Key Learning**
- Non-epileptic attacks reduce demands and anger.
- Being around others isn’t safe (continued non-epileptic attacks strengthen fear).
Discussion of Jayden’s MSFA. The hypothesised function of Jayden’s NEAD is related to a need to withdraw from difficult social and emotional situations, particularly situations which evoke anger. NEAs also increases the care he receives from his mother. The hypotheses developed from the following analysis.

Early experiences. The analysis of Jayden’s early childhood suggests that he had little opportunity to develop/learn adaptive strategies for dealing with and expressing emotions. Jayden learnt that withdrawing and keeping quiet was best, as being noticed by his drunk father would often lead to him being physically abused. Simultaneously, he learnt that illness behaviour and illness reporting increased care from his mother. As his father beat him in private, illness was also negatively reinforced; it served to reduce physical violence by being inadvertently protected by his mother.

Early adolescence and school. Faced with increased social demands as he began secondary school, and a lack of skills (due to childhood isolation and withdrawal), Jayden began illness reporting at school. This generalisation of operant behaviour was again negatively and positively reinforced, with him having time off school and being cared for by his mother on an intermittent reinforcement schedule (sometimes she was at work and could not leave). When unable to withdraw Jayden expressed his anger, in situations where he was bullied/teased he would respond with aggression/violence. This behaviour was negatively reinforced by reducing the bullying.

Head injury. Expression of emotion (anger/aggression) became positively reinforced, it was praised by his rugby coach and team-mates giving Jayden status at school for the first time. However, this behaviour was punished when Jayden experienced a head injury when he went in for an aggressive tackle during a match. The punishing consequences were immediate experiences of pain and disorientation. Jayden’s head injury, resulted in post head injury seizures (a respondent behaviour as they were unconditioned/automatic responses to organic stimuli). When respondent behaviours have reinforcing consequences they can become associated with the context (stimulus) in which they occur and be emitted in future, at which
point the behaviour is then under operant control. Though the seizures were uncomfortable, unpredictable, and embarrassing (aversive experiences), it is hypothesised that the reinforcing consequences were more powerful. The seizure behaviour was negatively reinforced due to reducing social demands (Jayden having time off sick from school). They were also positively reinforced by the increased care Jayden received from his mother. Jayden’s seizures were treated with anti-epileptic medication and it is hypothesised that this treatment reduced their frequency [see Critical Reflections for further information regarding this hypothesis, pg 143]. However, due to the established reinforcement of the behaviour, non-epileptic attacks which mirrored the seizures were emitted in context where seizure behaviour had been reinforced (non-epileptic attacks being operant behaviour, controlled by their environmental consequences). Additionally the non-epileptic attacks were less punishing than seizures as it appears Jayden remained continent during attacks but was not during seizures.

**Fight with father.** Jayden’s fight with his father served to punish his expression of anger further. Though Jayden reported hating his father and said he did not regret his actions, his father’s death is hypothesised to be punishing due to the fear of reprimand Jayden experienced. Jayden lied to his mother and others to avoid feared reprimand and an increase non-epileptic attacks served to avoid the difficult situations and to increase care (at a time he feared he may be rejected/punished).

**Restrictions on life.** In the longer term, Jayden became angry about the restrictions placed on his life by his epilepsy diagnosis. His efforts to lead an independent life were restricted by not being able to drive and attacks impacting his ability to work. Based on Jayden’s learning history it is hypothesised that when he found social situations difficult non-epileptic attacks were emitted which reduced these demands by enabling withdrawal. A further instance of Jayden being aggressive was negatively punished, when he threatened to punch his neurologist (who alluded to his attacks being “made up”), he was consequently discharged from the service. Though Jayden’s non-epileptic attacks were originally contingent on high social demands, when his anger was repeatedly punished, it became a similarly aversive stimulus. Through the
process of operant generalisation, influenced by Jayden’s severely limited behavioural repertoire, attacks were emitted and subsequently reinforced as Jayden’s anger decreased temporarily and feared punishment was avoided. Jayden’s continued attacks also elicited increased care from his mother. By this point she had learned of the physical abuse her husband had inflicted on Jayden and her increased caring behaviour was negatively reinforcing for her as it temporarily relieved her feelings of guilt. The fact that Jayden’s non-epileptic attacks sometimes resulted in injury to others and damage to property increased his anger but also led to him fearing being around others and strengthened the need to avoid. However, the most successful strategy for avoidance in his learning history were non-epileptic attacks. Further non-epileptic attacks emitted in response to anger (now it has been conditioned) seemed to serve to confirm his beliefs and strengthened his fear further.

Current context. It appears that Jayden continues to have some seizures which fit with epilepsy (in terms of semiology, and differing antecedents and consequences) though these appear to be much less frequent than hypothesised non-epileptic attacks. In his current life context the main anger inducing stimulus for Jayden is the demands placed on him by his partner. It is likely that a cycle of reciprocal reinforcement has developed within the relationship. In order to establish the relationship Jayden is likely to have responded to demands positively, but over time became less attentive (reinforcing the demand behaviour on an intermittent schedule). With the persisting demands Jayden’s anger increased and his attacks served to allow avoidance of the demands and he retreat to the safety of his mother’s care. Though his partner had wanted them to move in together Jayden continues to live with his mother and step-father. His fear of being around others appears to be particular pertinent regarding his daughter and he therefore lives with his mother and step-father rather than his partner. It appears that living separately exacerbates the demands made by his partner and thus a cycle seems to have developed.

Susan. Susan is a 62 year old, white female with diagnoses of NEAD, Functional Neurological Disorder (FND, described as generalised paralysis) and chronic pain. NEAD was diagnosed two years ago following lengthy
investigations after her reportedly long-standing somatic symptoms increased following a transient ischaemic attack (TIA or ‘mini stroke’) in 2004. She has been with her second husband for 20 years who she lives with and she has four adult children from her first marriage. She has seven grandchildren. Susan was recruited as she attends a neurology outpatient clinic in relation to her NEAD and FND.

Table 13. Susan’s MSFA

**Early experiences**

Susan, was the younger of two daughters by four years, brought up in a working class family with her mother and father. Her father had older children from a previous marriage who lived with their mother and her older sister was fathered by someone else before her parents got together. Her father worked when she was very young but experienced lots of illness during Susan’s childhood. Before Susan was born her mother lost an arm in an accident working in a factory (Susan’s sister was 2-3 years old) which meant she couldn’t work and it also impacted her ability to do the housework. As Susan’s mother had to work after having her first child (Susan’s sister) she blamed her for the injury and outwardly referred to it being her sisters fault. Susan described her mother as “bitter”, suggesting that they owed her for raising them and using her disability to make them feel guilty so they would do more chores.

From around three years old Susan recalled hearing her parents’ volatile arguments when she was in bed, with her mother being the main aggressor. She reported crying herself to sleep but eventually training herself to ‘be somewhere else’ in her mind to pretend it wasn’t happening. Crying in front of her mother was either ignored or elicited active dismissal (being pushed away and/or sent to bed), she was never hugged or given positive attention in response to a need or as praise. In light of her father’s health and her mother’s disability, Susan suggested that illness was not something that elicited extra care or attention in the family, “you just get on with it”. She reported having lots of illnesses including measles, German measles, and Scarlett fever in addition to frequent severe migraines and intense growing pains. She reported learning ‘not to make a noise when you cry’ as her mother noticing it would lead to being scorned, as it was regarded a sign of weakness.

Susan described her mother as the dominant parent and also an unstable woman. She recalled her mother threatening suicide on multiple occasions and threatening to leave with Susan’s sister (whom her husband was not the father to) leaving Susan and her father behind. Susan described her father as a good man, but that he was placid (termed “weak” by her mother). The whole family were fearful of her mother’s mood swings. She felt as though her mother’s behaviour sent the message “nobody should have an easy life”. Susan reported that her father was controlled by her mother and that he learnt not to show affection to them in front of her. In
her absence he would treat them to ice cream and allow them to sit on his knee but this was
often fleeting and marred by fear. Susan described her and her sister comforting each other but
that her mother caused arguments between them to stop them being close.

**Functional analysis sequence 1**

**Antecedents**

Susan observes older sister is punished for emotional expression, and for seeking
care/affection from mother.
Her father is also punished for emotional expression and submits to her mother.
Her parents argue violently at night.
There is a family history of ill health and disability.
Father is warm when mother is not there but cold when she is present.
Susan and her older sister have a good relationship.

**Behaviours**

**Covert**

Home is unsafe, I need to be on guard (fear)
If I express anything I am weak.
If I need others I will be punished.
I can be close to my father but this is risky (approach/avoidance conflict)
Distance self from painful present experience (dissociation when parents argue)

**Overt**

Limited reporting negative emotional experience to parents
Some reporting of emotions/distress when unwell
Submit to mother’s requests and general dominance
Seek comfort from sister

**Consequences**

Minimal positive interaction/attention with father.
Though Susan submits to mother, mother causes arguments between her and her sister to
interrupt their closeness
Emotional needs not met

**Key learning**

- Expressing negative emotions/distress will be punished.
- Even submitting doesn’t improve the situation (avoidance/avoidance conflict)
- Others can’t be relied on to be supportive and caring.
- Dissociation (going somewhere else) provides some relief from horrible situations
School and syncope incident
Susan reported being bullied throughout primary and secondary school by her peers based on her appearance, she cried in private but put a brave face on in public, walking away/avoiding them when possible. In one class in particular Susan was bullied in front of the teacher who did nothing, and at times Susan reported that the teacher teased her too. She reported that her few friends were quiet and not ones who would stand up to bullying. Instead of socialising with her peer group she spent time with girls older than her, and her older sister and her friends. During a running race organised by her sister and friends (which Susan reported she didn’t want to do but felt pressured to) Susan was coming last (which she put down to being overweight) and got upset about this wanting to cry. She crumbling to the ground coming round to those she was running with peering over her.

This event appeared to involve over-heating and panicked breathing, followed by the fall (a loss of consciousness involving losing the ability to perceive, process, and interact with her environment). This episode seemed to differ to Susan’s later descriptions of Non-epileptic attacks and it is hypothesised that this was an incidence of syncope (fainting). Susan reported having two similar episodes during school, in situations also involving physical exertion (running around the playground) but these were not associated with negative emotions.

Functional analysis sequence 2
Antecedents
Key learning from sequence 1
Susan is verbally bullied at school
Susan is pressurised into competing in a running race to please her sister and her friends.

Behaviours
Covert
I am unfairly treated, those who should help me don’t, I need to look after myself (anger and strengthening of rule governed behaviour)
I can’t look weak in front of my sister and her friends but I’m not good at this (conflict)

Overt
Susan hides her emotions and avoids bullies
Susan faints during the race

Consequences
Bullying remains constant (emotional suppression negatively reinforced as Susan believed bullying would have escalated if she had shown emotions).
Susan’s sister and friends don’t see her upset (fainting behaviour negatively reinforced similarly)
Susan’s sister and friends show concern for her (fainting behaviour positively reinforced).
Key Learning

- Hiding emotions and distress prevents being seen as/called weak.
- Fainting saved Susan from people seeing her as weak.

Onset of NEAD in the workplace and first marriage
Susan went to college to train in catering from the age of 16. Susan reported that she was considered a promising student and competent chef in the workplace. Within a male dominated environment Susan reports being verbally harassed/ridiculed by men (relating to her gender and her ability). Susan was hurt by their comments but did not show her emotions or “rise to it”. Susan came to perceive the bullying to be driven by jealousy. Though this hypothesis is particularly tentative due to limited data [see Discussion: Limitations], it appears that the onset of non-epileptic attacks may have been in such situations in Susan’s workplace. Susan reported having “blackouts” in the workplace and on one occasion she reported a particular link between increased emotions in response to bullying and having a “blackout”. The hypothesis is founded upon the slightly different semiology in terms of Susan reportedly retaining some perceptual ability typically hearing (though the non-epileptic attacks generally mirrored syncope).
Additional as the syncope was positively and negatively reinforced and Susan was unable to walk away (due to the risk of losing her job), it may be that non-epileptic attacks (based on the syncope behaviour) were emitted as the only other effective behaviour in her learning history. The NEAs appeared to elicit concern from colleagues and Susan developed a positive relationship with her family doctor through the lengthy and inconclusive investigations.

Susan met her first husband age 18 and married after three years together. She described him as being charming initially and in public, but that he quickly became controlling within the relationship. She reported that he disliked her working even though it was necessary financially, he was suspicious of her working with men, though she reports she was never unfaithful. Early in their marriage, Susan fell pregnant but suffered a miscarriage. Susan was physically and emotionally traumatised by this experience and as a consequence she was not interested in sex. When her husband initiated sex and she refused, he initially tried to persuade her but she was not comfortable with this. He ignored her wishes and forced himself on her. Susan reported that her first instinct was to fight but she was quickly overpowered and he raped her. Any time after this Susan refused sex he would rape her again. After fighting against him was not successful Susan reported adopting the strategy of going somewhere else in her head (dissociation), a strategy associated with night time in her childhood [see pg 135 for consideration of Susan’s reported dissociation].

Functional analysis sequence 3
Antecedents
Key learning from sequence 2
Working in catering, Susan’s ability is recognised and valued. Male colleagues verbally harass and ridicule Susan. Susan’s husband becomes controlling, questioning her and being suspicious of her working with men and having a social life. Susan has a miscarriage. Susan’s husband rapes her when she refuses to have sex with him.

**Behaviours**

**Covert**

I can’t let them see they have got to me (fear of emotional expression)

Susan dissociates during rape

**Overt**

Continue to go to work

Non-epileptic attacks begin at work

Reassures husband when accused

Compliance with husband’s controlling requests

**Consequences**

Non-epileptic attacks serve to suppress emotional expression (negatively reinforced due to feared consequences).

Non-epileptic attacks serve to increase the concern of others and investigations lead to a supportive relationship developing with her doctor (positively reinforced)

Marriage continues as does rape and physical abuse

**Key Learning**

- Non-epileptic attacks are effective in suppressing emotions
- Being ill means people are more caring
- Dissociating helps in horrible/terrifying situations

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**First marriage continued**

Susan had four children (through three pregnancies). Susan reported having what are hypothesised to be non-epileptic attacks during her pregnancies. However, as her sister had experienced ‘blackouts’ during pregnancy they were not treated with particular concern and were routinely investigated with no conclusion. Susan’s husband continued to rape her and he began to be physically violent also. This occurred in front of the children and he also began to be violent towards their son (the eldest child). In order to reduce the exposure to and impact of the violence on her children, Susan complied with her husband’s controlling demands and submitted to his physical and sexual violence (continuing to dissociate). She reported wanting to leave but having no other option financially. When her youngest child was two and her oldest was seven Susan had to go back to work as her husband developed a bad back and couldn’t
work so they needed the money. Susan reported continued non-epileptic attacks throughout the marriage though it appeared that over time Susan and others “got used to them”. Over the years Susan suspected her husband of having multiple affairs, and when her eldest child was 16, her husband said that he had been having an affair with a colleague for a year and he left the family home a short time later.

**Functional analysis sequence 4**

**Antecedents**
Key learning from sequence 3
Husband uses sexual and physical violence towards Susan, and is physically aggressive towards their son, and in front of the other children.

**Behaviours**

**Covert**
I am weak, I cannot protect myself or my children (powerless)
I don’t know what to do (sense of inertia due to lack of practical coping strategies)
Dissociation during sexual and physical violence

**Overt**
Continued non-epileptic attacks
Compliance with husband’s controlling requests – not working/working
Submission to violence in front of children

**Consequences**
The continued non-epileptic attacks were negatively reinforced (suppressing emotion)
Continued non-epileptic attacks elicited less concern from others (reduction in positive reinforcement)
Sexual and physical violence continues.
Eventually Susan’s husband admits to an affair with a colleague and leaves the family

**Key Learning**
- Non-epileptic attack continue to be effective in suppressing emotions
- Dissociating helps in horrendous situations

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**Transient Ischaemic Attack**
After her husband left Susan continued to work part-time to financially support herself and her children. Susan met her new partner (now her husband) reporting an opposite relationship to that with her first husband. Particularly she was considered the dominant one in the relationship. Susan worked in pubs for a while but ended up working in a restaurant kitchen which she really enjoyed to begin with. Susan reported being good at the job (having trained in catering) and she was quickly given more responsibility.
In 2004 Susan had several workplace conflicts with colleagues (who Susan reported were jealous of her quick progression), and management, who she reported were increasingly demanding but also patronising. Her children were more demanding of support due to having their own issues; her daughter developed anorexia, her son was taking drugs and another daughter was diagnosed with epilepsy. Around this time Susan experienced what was later confirmed to be a Transient Ischaemic Attack (TIA, or mini-stroke) whilst at work. Susan experienced loss of sensation in her right side and a ‘bang’ in her eyes resulting in tunnel vision. Susan took a short break but came back to finish her shift, she reported that she knew something was wrong when she couldn’t pick something heavy up but her shift soon ended. Susan went back to work for her next shift (Monday), during the shift she was shaking and felt sick which was followed by a non-epileptic attack. She went to hospital following this though no explanation was found for the “funny turn”. She had a few days off work but then returned. In addition to continued non-epileptic attacks Susan experienced extreme tiredness and pain and difficulty concentrating. Her GP signed her off sick for a month, and as her symptoms continued she continued to receive sick notes. After being off sick for around a year she quit work. Medical investigations continued and around two years later it was confirmed that she had experienced a TIA.

**Functional analysis sequence 5**

**Antecedents**
- Key learning from sequence 4
- Positive relationship with second husband
- Children are facing own life issues and needing increased support from Susan
- Work is stressful
- Susan experiences a TIA.

**Behaviours**

**Covert**
- Everything is stressful, nothing is ever easy for me (anger)
- I feel guilty for my children’s problems because I didn’t protect them from their father (guilt/pressure to act)
- I cannot cope, but I need to be able to or people will think I am weak (fear of emotional expression)

**Overt**
- Non-epileptic attacks increase
- Susan experiences fatigue and pain and has difficulty concentrating.

**Consequences**
- People are increasingly concerned about Susan and there are significant medical investigations through which Susan receives renewed support from her GP (attacks and other symptoms
positively reinforced)
Non-epileptic attacks continue to suppress emotions (negatively reinforced)
Susan is signed off work sick and eventually leaves (NEA’s negatively reinforced through avoidance of stress/emotion inducing stimuli)

Key Learning
- It is more difficult to cope since having the TIA
- Being stressed seems to lead to NEAs (“blackouts”)

Family stress and current context
Following quitting work Susan experienced varying levels of stress, usually related to her children having difficult times and needing her/asking her to support them. Susan reported finding it difficult to support them in light of her continued symptoms. Furthermore, following her diagnosis of NEAD two years ago Susan’s belief that stress could lead to an attack was confirmed. Susan reported that it was them more important that her family recognised this and supported her. She feels that her children don’t understand the seriousness of her illnesses and that they don’t demand less of her as a result. Susan reported that she wants to help but that she is very worried about her ability to deal with the stress in light of her illnesses. Susan experienced increased non-epileptic attacks when her husband’s health was compromised (following a heart attack) and when her benefits were under review.

Within the interviews for this research, Susan had non-epileptic attacks on three occasions. The first time was when Susan was explaining how the researcher should act if she were to have an attack. The other times were when Susan was describing current sources of stress.

Functional analysis sequence 6
Antecedents
Key learning from sequence 5
Life stress including: Susan’s daughter’s eating disorder becomes serious, Susan’s husband becoming unwell, Susan’s benefits being reviewed

Susan is asked to describe how the researcher should react should she have an attack
Susan is asked to talk about current sources of stress in her life

Behaviours
Covert
I can’t cope with stress, my body can’t cope, I will have an attack (anticipation)
They should understand I can’t deal with this (anger/upset)
They are not taking my illnesses seriously (anger/upset)
I’m going to have an attack and she won’t know how to react (fear/anticipatory embarrassment)
I can’t cope with stress, my body can’t cope (generalisation to fear of emotional experience due
to anticipating attacks)

**Overt**

Non-epileptic attacks

**Consequences**

Stress and demands are temporarily reduced (NEAs negatively reinforced)

Susan's fear/anticipation of NEAs is reinforced.

Susan doesn’t have to continue thinking about the stress and intolerable thoughts for a short time (NEAs in response to thoughts negatively reinforced – generalisation)

**Key Learning**

- Stress should be avoided.
- Sometimes attacks have no specific trigger – they can happen at any time (Susan doesn’t recognise covert behaviour as a stimuli, only external sources).

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**Discussion of Susan’s MSFA.**

**Early experiences.** Susan witnessed and experienced negative emotional expression to be punished by her mother. When she cried or expressed distress her mother would ignore her or actively punish her as she got older (sending her to bed or pushing her away). In the presence of Susan’s mother her father would ignore her but when her mother was absent he would offer care and love. This punishment and approach/avoidance conflict led to a reduction in emotional expression behaviour. Due to emotional experience being an unconditioned response Susan had to learn how to suppress her expression of emotion. Susan learned to avoid situations or if this was not possible (as in her home environment) she began to dissociate from the current emotion-evoking situations. This behaviour was negatively reinforced as it meant she avoided being punished.

**School and social life.** Susan reluctantly participated in a running race with her sister and friends. She was losing, struggling to run and feeling upset about coming last. Being hot, sweaty, tearful and hyperventilating (over-breathing), Susan fainted. It is hypothesised that Susan fainted (organically rather than psychologically underpinned) due to her pre-faint behaviour, descriptions of ‘collapsing’, being ‘out’ for a few seconds, and not recalling any sensory perception whilst she was out (which contrasts with her later
descriptions of NEAs). Fainting meant Susan was able to avoid negative emotional expression which she anticipated would have led to being punished/her being judged as weak (based on her previous learning at home). The fainting was therefore negatively reinforced and through this experience Susan’s belief was not disconfirmed. Additionally the fainting behaviour was positively reinforced as her sister and friends ran to her aid and offered care and attention to Susan (whereas illness/injury had been largely ignored previously). When Susan was verbally bullied at school (most significantly in secondary school) her beliefs remained that expressing her emotions would lead to punishment (bullying getting worse) and being considered weak (something she had learned was the worst possible thing to be). Though she experienced negative emotions in response to bullying, she suppressed these or withdrew from the situations. Susan believed that the bullying remaining constant/not getting worse was due to her emotional suppression (mirroring her relationship with her mother).

**Early work experience.** In the workplace Susan was bullied by male colleagues. Susan felt that this bullying was driven by jealousy due to her natural ability in the role. It is hypothesised that this perception (which may well be accurate) may have served as a protective mechanism to enable Susan to suppress her emotions in an environment where withdrawing would have negative consequences (losing her job). However, it seems that the onset of non-epileptic attacks may have been in such situations. On one occasion in particular Susan recalled a link between increased emotions in response to bullying and having a “blackout”. It appears that as the syncope was particularly effective in suppressing emotions and eliciting care in a social context, and Susan became particularly emotional, a non-epileptic attack mirroring syncope was emitted. The NEA was effective in suppressing emotional expression and was additionally positively reinforced by eliciting concern from colleagues and sparking the development of a supportive relationship between Susan and her family doctor.

**First marriage.** Susan’s first husband was initially charming but quickly became controlling of Susan in private. Susan had learned to submit to the controlling behaviour of others by witnessing her father’s submission to her
mother. Though Susan had no expectation that submission would have positive consequences based on her childhood experiences it was the behaviour she had learnt in response to dominant others. Susan being raped by her husband following a miscarriage led to dissociation, the childhood strategy in response to fear in a similarly private context) this was reinforced as Susan was able to partially avoid the current traumatic and painful experience. Though the two behaviours in this life stage were different, it is hypothesised that they had similar functions. Dissociation was associated with night time emotional suppression and cognitive avoidance of aversive experiences and non-epileptic attacks served to suppress emotional expression which Susan feared would be punished/perceived as weak, in her daily life.

**First marriage continued.** Susan’s husband continued to rape her and also began being physically violent towards her in front of their children, and towards their son. By this time in her life Susan had learnt few effective strategies for escaping from aversive situations and suppressing her emotions, her limited repertoire consisted of withdrawal (less well established due to aversive home environment in childhood), dissociation, and non-epileptic attacks. Withdrawal was less well established and was also a less feasible option due to the need to financially support her children. Dissociation and non-epileptic attacks continued to be effective and therefore reinforcing in the short-term, she was able to ‘escape’ temporarily. It is hypothesised that Susan’s behavioural repertoire was so limited she was almost in a position of learnt inertia. Behaviours other than complying with the requests of others had been punished up to this point, and the behaviour with positive consequences (dissociation/non-epileptic attacks), had little active impact on others. Susan had learnt that she had little control over her environment and her life. This is exemplified by the fact that the abusive relationship with her husband only came to an end when he left Susan and the children for another woman. As people got used to her non-epileptic attacks they appeared to elicit less concern/support and were less positively reinforced, though negative reinforcement continued.

**Transient Ischaemic Attack.** Susan had met her new partner, her children were now young adults, and she was working part-time. Susan
continued to experience non-epileptic attacks in response to emotional situations and as she did not express her emotions her belief learnt in childhood was never disconfirmed. When Susan's life became more stressful, in the workplace and in the family, Susan experienced a Transient Ischaemic Attack (mini-stroke). When Susan's non-epileptic attacks increased in frequency in response to situations evoking negative emotions, they were deemed to be part of her ‘post-stroke symptoms’. Within this milieu they received renewed attention and investigation, and support from her doctor. The short-term negative reinforcement of avoiding emotions continued and during this time she was signed off work sick (NEAs were also negatively reinforced by reducing exposure to stressful stimuli). With the increased frequency, and investigations, for the first time Susan noticed an association between stressful situations and the non-epileptic attacks.

**Family stress and current context.** Recognising the link between stress and attacks, Susan expected that others would reduce their expectations of/demands on her, including her children. After the initial supportive response to her stroke and symptoms waned, Susan was expected to revert back to supporting others which led to her beginning to feel angry towards them. This anger increased when stress was confirmed as a potential trigger when she was diagnosed with NEAD. Susan’s attacks continued in response to emotional and stressful situations (increasing in frequency at particularly stressful times). Susan’s attacks also began occurring in anticipation of stress and in response to her anger towards others regarding their lack of continued support and re-emerging demands. This was evident in our interviews, when Susan was talking about current stress and the expectations of others, she had non-epileptic attacks on two occasions. In contrast, talking about past traumatic/emotional/stressful experiences Susan did not have any non-epileptic attacks. This suggests that non-epileptic attacks are a behaviour contingent on the discriminative stimulus of current stress, memories and related emotions regarding the past do not appear to have been conditioned as stimulus for non-epileptic attacks. Additionally, Susan had an attack when she was explaining to me how I should react if she had an attack during the interviews. Susan later described the worst thing about the attacks as her fear of how people might
react, and that it might be embarrassing. Susan reported having attacks when people are investigating her illnesses. It appears that this anticipatory anger, fear and embarrassment are generalised stimuli for non-epileptic attacks. Susan no longer needs to be exposed to an external stressful/emotional stimuli for her to have an attack. Susan's thoughts which evoke emotions also lead to attacks, which similarly temporarily suppresses her emotions and feared emotional expression and she avoids the stimuli (in this case her thoughts). The consequence of this generalisation is that Susan thinks her blackouts have no specific trigger at times, she continues to look for external sources of stress/emotion neglecting to recognise her internal experiences as possible triggers.

**Daisy.** The analysis presented in Table 14 is the MSFA for Daisy. Daisy is a 31 year old, white female with diagnoses of NEAD, fibromyalgia, and myalgic encephalopathy. NEAD was diagnosed around two years ago, whereas the other diagnoses were made shortly after she experienced what was initially suspected to be a stroke (later diagnosed as functional hemiparesis) in 2009. She has been with her husband for 10 years and together they have two children. Daisy was recruited as she attends a neurology outpatient clinic in relation to her diagnoses.

Table 14. Daisy’s MSFA

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**Childhood**

Daisy, has an older full-brother and two younger half-brothers. Her mother and biological father’s relationship was violent, he violently beat her and the children (but mainly Daisy’s brother). After a particularly severe beating, Daisy’s mother left her father with her and her brother, Daisy was four at the time. Her mother soon met Daisy’s step-father who she is still married to. Being brought up in this household Daisy described a home where all of the children were expected to contribute to the household chores and they were all encouraged to be independent. Daisy described her step-father as “regimented” wanting jobs done in a certain way, and he expected children to be seen and not heard. Though both parents worked, this was not always the case and they spent times struggling financially. It seemed to Daisy that when they worked they would spend a lot (even beyond their means getting into debt) and particularly her mother wanted the best things, which made her happy. She reports that her mother was loving but preoccupied with her own life, particularly with “keeping up with the Joneses”. Her mother also reportedly experienced some mental health difficulties, Daisy reported that her
mother was "unstable" and both her mother and step-father were on anti-depressants. In this relationship she recalled her mother being violent to her step-father from early in their relationship. At various points in her childhood her mother would react to arguments with her husband by taking the children away for a night to friend's houses (though her full-brother, two years her senior, over time began to refuse to go). Daisy recalled becoming used to this and recognising the pattern that they would soon be home so she didn't worry or become upset. Daisy recalled her step-father making a suicide attempt following one of the arguments, which her mother was open with all of the children about. Daisy feels that she was closest to her youngest half-brother who is 13 years her junior, feeling as though she took on a motherly role with him, by this point realising that her mother was "not good enough". Daisy was expected to look after her half-brothers whilst her parents worked, she would never say no.

**Functional analysis sequence 1**

**Antecedents**
Daisy’s parents are often preoccupied with their mental health issues, their conflicts, or portraying a positive image to others through spending money.
Daisy and her siblings are expected to do household chores, be obedient and be quiet.
Daisy is expected to look after her younger siblings.

**Behaviours**

**Covert**
Feeling valued when she is productive
My mother isn’t good enough, when I grow up I don’t want to be like her

**Overt**
Daisy cares for younger siblings.
Daisy completes household chores.

**Consequences**
Caring behaviour is positively reinforced by the relationships developed with siblings and negatively reinforced because it helps keeps the home harmonious.
Caring for her siblings also allows her parents to work and earn more money to buy nice things (positively reinforced).

**Key Learning**
- Having nice things/money is important for being happy.
- Others’ needs are more important.

**School, early career and fainting incident**
Daisy reported working hard at school, enjoying it but recognising that she needed to try more than some others to get good grades. Daisy reported liking school and preferring it to being at
home. Daisy began part-time work aged 13 which was encouraged by her mother. Daisy reported enjoying having money to buy nice things for herself. Academic achievement was important to her parents and became important to her, Daisy felt as though her mother had high expectations and piled pressure on her to do better than her older brother had to “redeem” the family (he performed badly). Daisy experienced migraines whilst studying hard for her exams. When her mother took her to the GP they were suggested to be stress related, an explanation which was acceptable to them both. Daisy took standard painkillers but continued to work hard as before. When she was successful in her GCSEs (achieving B’s and C’s) Daisy recalled being given a camera as a present from her parents, which was left on her bed. She described it as the first big gift she got, particularly compared to her half-brothers who were favoured (most notably they were taken on holiday to Florida whilst a 16 year old Daisy was left at home and not offered the chance to go). She described resenting having to buy her own uniform for sixth form college, though she enjoyed the independence of working.

After leaving sixth form, Daisy worked full-time. Daisy began dating her now husband when she was 20 years old. As well as full-time work she had taken on an evening job in a pub which she reported was a stressful and tiring time. Though her primary job was “well enough paid”, Daisy wanted to earn more to buy nice things. During this time Daisy caught a virus and became unwell, she was made to go home from working in the pub early one night due to being unwell and Daisy reported that she “blacked out” in her bedroom after getting home. Daisy went to hospital and the virus was detected/confirmed, as a result she spent two weeks off work recovering in bed. Daisy returned to work, she soon became pregnant with her first child. Within a few years Daisy gave birth to her second child. She began working in insurance claims and was promoted to a supervisory role within a year. Daisy got married when she was 25, as a couple they were earning enough to buy everything they needed and wanted. In the house Daisy was in charge of everything (which was her choice), she describes herself cleaning excessively and wanting everything to be perfect.

**Functional analysis sequence 2**

**Antecedents**

Key learning from sequence 1

Daisy’s mother pressurises her to perform well at school to do better than her brother
Daisy’s mother encourages her to work and she is soon expected to pay for her own clothes including her uniform
Daisy becomes unwell with a virus whilst working a full-time job and an evening job.

**Behaviours**

**Covert**

I enjoy being independent but I am jealous that my siblings get more than me
I want to do well then I can buy myself the things I want on my own (determination)
Even though I’m unwell I can’t afford to have time off
Overt
Daisy works hard at school
Daisy works hard in part-time job
Daisy experiences migraines whilst studying
Daisy faints after being sent home from work early due to being particularly unwell

Consequences
Working hard is positively reinforced with money from job and camera from parents
Migraines are investigated and treated with painkillers
Daisy is taken to hospital by her boyfriend (now husband) and has two weeks off to recover
(fainting is negative reinforced through reducing stress and enabling recovery from the virus).

Key Learning
• Working hard must be prioritised, others expect this and it is beneficial for everyone
• Fainting meant Daisy had time off work, stress reduced and she recovered from the virus

Functional hemiparesis
Daisy reported juggling full-time work, keeping the house immaculate, and raising her two children (her husband was happy to help but she wanted to do it all). In the week before her youngest daughter’s birthday party Daisy experienced a severe and prolonged migraine (worse than any she experienced at school), she took painkillers and continued with the work/plans. During the party Daisy experienced intense pain in the right sight of her face and neck, she drunk wine and carried on. When Daisy’s migraines returned and painkillers were ineffective, Daisy was directed to the hospital, where they suggested she was experiencing severe migraines. Daisy reported that at work three weeks later she experienced loss of sensation in her right side; she was unable pick up paper with her right hand. Her husband picked her up from work and took her to the hospital. Daisy spent five days in the in-patient stroke unit and it was concluded that she had experienced a functional stroke resulting in functional hemiparesis.
Daisy reported that she experienced continued symptoms including severe migraines, generalised pain, and fatigue (which after around six months were diagnosed as Fibromyalgia and Myalgic Encephalopathy by her Consultant Neurologist). Around nine months after the stroke Daisy went back to work part-time on administrative duties. Daisy reported continuing to work and run the house (reporting that she wouldn’t let anyone else do it). One-two years after returning to work Daisy’s manager put pressure on her to return to her previous role part-time, which Daisy agreed to do. Daisy reported that she was “getting by, going through the motions”, she came home from work and went straight to sleep. She could no longer manage the housework but Daisy reported that she continued to function in a “zombie like” state for over a year.
Functional analysis sequence 3

Antecedents
Key learning from sequence 2
Daisy puts herself under pressure to manage full-time work, raise her two young daughters, and keep the house immaculate
Under increased stress Daisy experiences symptoms which are diagnosed as a functional stroke leading to functional hemiparesis (weakness)

Behaviours
Covert
I have to make sure the girls get everything they want, so they don’t feel like I did
I can’t let people down (through learning to put others’ needs first)
Overt
Daisy experiences continued migraines, fatigue and pain
Daisy returns to work on light duties
Daisy continues with housework
Daisy returns to her previous role

Consequences
Daisy’s return to work is positively reinforced, with money and with praise
Daisy is too tired to do housework after being at work

Key Learning
- Meeting the needs of others and doing what they expect is all that matters

Non-epileptic attacks
Daisy continued to work despite continuing to experience migraines, pain and fatigue. Daisy reported that expectations and demands increased over time. When Daisy’s manager went on holiday Daisy was expected to take on (and did take on) increased responsibilities. Daisy reported that this time was extremely stressful and she was experiencing the most severe symptoms particularly migraines. After coming home from work one day Daisy experienced what is hypothesised to be her first non-epileptic attack. Daisy reported being in the kitchen trying to clear up (notably doing housework was now rare due to her symptoms) when she felt her vision go blurry. Daisy recalled trying to hold onto the sideboard but falling to the floor, banging her back on a plug when she fell. Daisy’s oldest daughter was there and telephoned Daisy’s husband. It appeared that Daisy was only ‘out’ for a few seconds, she reported feeling very disorientated when she came round. Due to the injury to her back Daisy had a few days off work following this. The next hypothesised non-epileptic attack occurred when Daisy had returned to work and was preparing for an important meeting. Daisy reported feeling extremely
stressed which was made worse by a disagreement with a colleague she had an on-going difficult relationship. Daisy went to toilets as she felt unwell, and in the toilet cubicle Daisy had the non-epileptic attack. Daisy reported coming round after what must have only been a few seconds, she recalls being aware of what had happened but she couldn’t get up straight away. Daisy went back to her desk and emailed her husband who collected her from work and she went home to bed. The next day Daisy telephoned her Consultant Neurologist who said he thought these episodes were ‘blackouts’, she was signed off sick for two weeks. Daisy went back to work but soon after she had another non-epileptic attack, though this was at home after work. Daisy reported that she had a “breakdown” when driving to work one morning soon after, breaking down in floods of tears. Daisy went off sick again and coincidentally within a few days she had her routine outpatient neurology appointment. Daisy's Consultant Neurologist warned her that if she continued to work she would end up back in hospital. The Consultant Neurologist called the blackouts non-epileptic attacks during this appointment and advised Daisy not to drive. Daisy reported that this made her fearful, she re-evaluated her belief that being off work was only temporary and she quit work. Daisy received the official clinical letter which included the diagnosis of NEAD.

**Functional analysis sequence 4**

**Antecedents**

Key learning from sequence 3

Daisy is put under increased pressure at work

**Behaviours**

**Covert**

I need to do this but I am struggling to cope (extreme stress).

**Overt**

Non-epileptic attacks

**Consequences**

Daisy has time off work following attacks (NEAs negatively reinforced)

Daisy experienced some injuries from attacks and her children became upset (NEAs positively punished)

Daisy is confronted by her Consultant Neurologist that she may end up back in hospital

Daisy quits work

Daisy is diagnosed with NEAD and is advised not to drive by her neurologist

**Key Learning**

- Non-epileptic attacks lead to rest, reducing stress and symptoms
- Quitting work will mean things get better.
Adjusting to limitations

After quitting work Daisy reported that she increased the amount of housework she did “to compensate” and “didn’t really slow down”. In the months after quitting work Daisy experienced five further non-epileptic attacks. With these occurring at home Daisy became increasingly concerned as her children were witnessing her attacks, and her youngest child was particularly upset by them. Additionally as the attacks continued, Daisy became more concerned about ending up in hospital as she had been warned. Daisy reported that she began to recognise that the attacks were always preceded by migraines, though not all migraines were followed by a non-epileptic attack. Instead of carrying on Daisy began to respond to increasingly painful migraines by going to bed/resting. Daisy also started pacing herself when she was feeling ok, she reported recognising that if she did too much on a good day she would have more symptoms the next day. Daisy reported that she also began to accept more help from her husband and became used to the house not being immaculate, feeling less anxious about it as time progressed. As Daisy was no longer working the family had less money, Daisy began to accept not being able to buy everything the girls wanted and everything she wanted for them. Since the cluster of eight NEAs in the first six months Daisy has had only three further NEAs in the last 18 months. Daisy recognises that these have been during times of particular stress and when she has become complacent “doing too much”.

*Functional analysis sequence 5*

**Antecedents**

Daisy experiences five more attacks in the months after quitting work which are witnessed by her children who become increasing scared and fearful (attacks positive punishment increased)

**Behaviours**

**Covert**

My children shouldn’t be exposed to this (their needs are most important).

It seems that bad migraines lead to me having attacks.

I can’t do what I used to, so I need to do things differently (further re-evaluation).

**Overt**

Daisy starts responding to migraines by going to bed.

Generally Daisy reduces the amount of housework she does.

**Consequences**

Daisy’s non-epileptic attacks reduce significantly (only three more in 18 months).

Daisy’s other symptoms become less severe (though still debilitating) (going to bed/doing less is negatively reinforced).

Daisy’s children are less scared (doing less-going to bed is negatively reinforced).
Key Learning

- Being healthy is more important than being able to buy things and clean the house.
- Self-care is important, it helps everyone.

Discussion of Daisy’s MSFA.

*Early experiences.* The key early experiences hypothesised to relate to later NEAD development concern a lack of learnt coping strategies. Daisy functioned well in her childhood in a strict environment. She learnt that academic success and general performance is rewarded. It appears that Daisy dealt with negative emotional experiences by focusing on other things, doing the things she knew would be rewarding for herself and rewarded by others. She also began to develop her caring behaviour (putting other’s needs first), through looking after her younger half-brothers. Initially this was requested by her parents and it was negatively reinforced by reducing household stress (enabling her parents to work), however, it was also positively reinforced by the relationship she developed with them, coming to see herself as a better mother than her own.

*School, career and fainting incident.* Daisy’s focus on working hard to be rewarded continued by taking on part-time work as a teenager and doing well in her exams. This general ‘working hard’ behaviour was rewarded with material possessions and was encouraged by her mother’s need to ‘keep up appearances’ and ‘look good’. In her early work life, Daisy had met her partner (now husband) and she was working two jobs; a full-time day job and a bar job in the evenings. Daisy’s working hard behaviour persisted and though she reported that it was very tiring and stressful it was positively reinforced through earning extra money to buy herself nice clothes and other material possessions. Whilst working these two jobs Daisy became unwell, she was made to leave her pub job early one evening because of this and when she got back home she fainted. Daisy went to the hospital and it was found that she had a viral infection and fainting was related to this. At this point Daisy had two weeks off work recovering from the virus, spending most of the time in bed, returning to work after this recovery period. This respondent behaviour (occurring automatically)
was negatively reinforced as Daisy’s stress reduced during that time and she recovered from the virus.

In the next few years Daisy had her two children, married her partner and began working in a new job with a better salary and more responsibility. Daisy worked full-time, kept a pristine house, and looked after her young children and at this time she felt this was necessary and it was achievable. By this time Daisy’s caring behaviour had become well established, she was used to doing everything and liked it this way. Working hard, caring for others, and keeping the house clean was reinforced by a sense of achievement in herself (influenced by her early experiences), the monetary reward, and by praise from others.

**Functional hemiparesis (weakness).** Daisy began to experience stress and tiredness managing work and home life. She had been promoted at work and was under more pressure as a result. Additionally looking after two young children had increased her stress. In the days before her daughter’s birthday party Daisy experienced severe migraines, when painkillers didn’t work she went to hospital it was confirmed to be nothing more than serious migraines. Daisy continued as before, working hard and putting other’s needs before her own (despite the continued symptoms). A few weeks later her weakness symptoms increased in severity and she was admitted to the stroke unit in hospital. Daisy spent five days there and was sent home on medication for migraines with a diagnosis of functional hemiparesis. The consequence of this episode of illness was that Daisy was off work for around nine months with continued symptoms (diagnosed as Fibromyalgia and Myalgic Encephalopathy six months after leaving hospital). During this time Daisy continued to do the housework and look after the children to her previous standards which continued to be positively reinforced, despite continuing symptoms. After nine months Daisy returned to work part-time in a less demanding role. It is hypothesised that although the illness was negatively reinforced through reducing stress having time off, this was not as reinforcing for Daisy as it might have been for others, she wanted to perform/work based on her previous learning of its importance and reinforcing value. Daisy was put under pressure by her manager to return to her previous role though still part-time. Putting
other’s needs before her own and having a learning history that this behaviour was rewarded, Daisy agreed. Daisy struggled to achieve what she used to be able to though the positive reinforcement of praise and money kept Daisy engaging in the behaviour. Daisy’s work behaviour appeared to be more reinforcing than her house keeping behaviour (influenced by Daisy’s learning and prioritisation of financial rewards) as during this time she began to do less housework and slept a lot at home in order to continue working.

**Non-epileptic attacks.** Though sleeping and doing less housework appeared to reduce stress for Daisy temporarily, she was put under increased stress when she agreed to take on extra responsibilities whilst her boss was on holiday. These levels of stress in conjunction with her physical symptoms (weakness, pain, and fatigue), appeared to mirror the context when she was 20 years old, working two jobs with a virus. In this context, and due to a severely limited behavioural repertoire, it is hypothesised that non-epileptic attacks (mirroring fainting) were emitted as a response to the similar stimuli (through the process of stimulus generalisation). The non-epileptic attacks were reinforced by leading to short-term reductions in stress and symptoms (e.g. the rest of the day in bed or a few days off sick). The attacks continued as they had been reinforced and eventually Daisy was confronted with the possibility of ending up back in hospital by her Consultant Neurologist leading to Daisy giving up work.

**Adjusting to limitations.** Though she had left work Daisy resumed her housekeeping and caring behaviour to regain the positive reinforcement she was lacking (compensatory behaviour). Daisy’s migraines, pain and fatigue symptoms increased leading to increased stress and worry (that she may end up in hospital). Daisy experienced further non-epileptic attacks. Though the negative reinforcement remained (resting and reductions in stress and symptoms), the attacks also had punishing consequences. Injuries Daisy sustained and the impact of witnessing the attacks on her young children acted as positive punishers for the behaviour. With Daisy’s previous learning of putting others’ needs before her own and her love for her children, the negative impact on her children (them screaming/crying and talking about it) had significant punishing value. It is hypothesised that this punishment outweighed the negative reinforcement of short-term respite for Daisy. With more
experience of attacks Daisy noticed an association between severe migraines and non-epileptic attacks. Recognising this and taking advice from her Consultant Neurologist Daisy engaged in a new behaviour, going to bed/resting in response to severe migraines. In this way the migraines served the function of eliciting rest/reduction in activity and so the NEAs were no longer required to meet this need. Learning that this was effective in reducing her symptoms, taking the professional advice, and having less time due to resting, Daisy also began to reduce her activity (including housework). Consequently these new behaviours reduced NEAs (and therefore the punishment of injuries and her children’s reactions). Daisy was attack-free for a year, however in the last six months she has experienced three NEAS. The functional analysis, and Daisy’s own insight suggested there are similar antecedents, Daisy being more “complacent”, doing more housework than she should and also a more stressful context (typically relating to financial troubles).

**Discussion**

This section of the thesis discusses the similarities and differences between the three MSFAs, and their relation to existing theory and research evidence. The limitations of the study are presented, clinical and research implications are suggested and finally my critical reflections including ethical issues are detailed.

Research into NEAD has identified psychosocial factors relatively common in this heterogeneous population. Such evidence, gathered primarily from correlational studies, demonstrates the presence of traits and experiences in NEAD patients, though the underpinning processes have not been thoroughly examined. The analyses presented above suggest that, at least for these three individuals, the aetiology and maintenance of NEAD can be understood functionally.

**Are there similar pathways in the development of NEAD for the different individuals?** Whilst the above analyses present the development of non-epileptic attacks in the three participant’s histories, an across-case analysis allows consideration of the similarities and differences in the developmental pathways.
The behavioural concept of symptom modelling was relevant in all the individual’s development of NEAD (Hopkins, 1989; Ramani et al., 1980). Though Bautista, Gonzales-Salazer and Ochoa (2008) suggested an extension of symptom modelling to include observation of seizures, here the direct (not observed) experience of other altered states of consciousness (syncope), are also hypothesised to be key in the development of NEAD. Each participant’s non-epileptic attacks were topographically similar to earlier seemingly organically underpinned altered states of consciousness (seizures or syncope). Jayden’s NEAs were similar though not identical to post-head injury seizures, Susan’s NEAs mirrored an incident of syncope in her childhood, and Daisy’s NEAs resembled the syncope she experienced when suffering with a virus. Whilst, Benbadis and Chichkova (2006) proposed psychogenic pseudosyncope as a provable and distinct diagnosis, Susan and Daisy attracted NEAD diagnoses. As the development and hypothesised functions were found to be similar for Jayden and Susan, a functional and formulation based approach (with the potential to inform treatment) seems more useful than suggested diagnoses/categories based on differing semiology.

The context and consequences of these earlier altered states of consciousness stand out as relatively unique instances, within difficult life circumstances, where the participants were relieved of pressure, or received support for the first time. It is hypothesised that as their lives progressed, in similar contexts (antecedents), this altered state of consciousness behaviour, may have been the only learned behaviour within their repertoire that would lead to successful/ameliorative consequences. Reuber (2009) suggested that an initial incident of syncope may increase sensitisation to and subsequent avoidance of similar pre-syncopal symptoms which are commonly experienced (theorising the role of panic/hypervigilance in NEAD development). In these cases however, the initial altered state of consciousness seemed to have overall positive consequences for the individuals, otherwise according to behavioural theory, the behaviour would be less likely to occur in future.

Jayden’s NEAs appeared to develop alongside his post-head injury seizures as they were resolved or successfully treated. In contrast to Susan and Daisy, Jayden’s experience of altered consciousness was not a one-off. Immediately
following his head injury Jayden was frequently experiencing seemingly organic seizures. It is hypothesised that the learning during this period of time led to the onset of NEAD as the seizures reduced in frequency (either due to successful treatment with medication or natural resolution as the brain healed). Susan’s onset of NEAD was less clearly hypothesised due to limited information available, though it seems that the first emergence of NEAs was nearly ten years after the syncope incident. This was when Susan was around 17 years old, being bullied in the workplace with continued fear of negative consequences for expressing emotion. The onset of NEAD for Daisy was around nine years after her virus-induced syncope, when she was experiencing physical symptoms and was under similarly significant stress.

Both Jayden and Susan’s NEAs generalised as they continued. Jayden’s attacks appeared to be originally contingent on social demands. When his anger also became an aversive experience (due to its negative consequences), through the process of operant generalisation and in light of Jayden’s severely limited behavioural repertoire, attacks became a response to his experiences of anger. Furthermore, his fear of the negative consequences of having attacks elicited anger and as such an explicit external source of anger is not always necessary for an attack to be emitted. Originally Susan’s attacks appeared to be a response to negative emotional experience and the learned need to suppress emotional expression, through actual punishment initially and a persisting fear of punishment. Susan appeared to become sensitised to NEA-inducing stimuli, over time her anticipation of attacks and thoughts about current situations which upset or angered her also preceded attacks. This suggested mechanism also provides an explanation for the ‘suggestibility’ of NEAD patients to induction techniques. To facilitate diagnosis Susan’s Consultant Neurologist induced an NEA using hyperventilation (Benbadis et al., 2000) in order to assess memory recall (LaFrance et al., 2013). The hypothesis that Susan’s NEAs became generalised to be emitted in response to internal stimuli (anticipation and thoughts) would explain her having an NEA in this situation with no external source of stress.

In contrast, Daisy’s NEA frequency reduced considerably since onset (eight in the initial six months, one year attack-free and then three in the last six months).
For Daisy the punishing consequences of the attacks (injuries, her children being affected, and continued functional symptoms), outweighed the positive short term consequences of reduced symptoms through enforced resting. Daisy felt that the diagnosis and further input from her consultant neurologist were key in making behavioural changes.

The difference between improved and continued NEAD in these three cases appears to be that attacks continued in cases where positive consequences continued in the context of difficult life and emotional experiences. Once Daisy had quit work and her attacks didn’t improve, fear of ending up in hospital and the impact on her children as they continued to witness attacks outweighed the reinforcement of attacks. It is hypothesised that by this time she was more likely to take on board advice/new behavioural strategies due to the current behaviour (attacks) being punishing, and the lack of other options in her learning history.

This may relate to ordering of treatment, with it necessary to modify (or at least highlight) attack reinforcement contingencies before problem solving and advising/teaching new behaviours.

Moore and colleagues (1994) considered NEAs as a tool to manipulate an individual's life more to their liking. However, upon finding higher levels of depression in NEAD patients compared to epilepsy and healthy controls, they proposed it would be an inefficient method unless the individuals would be in a worse position without the attacks. They suggested attacks may offer immediate/short-term benefits, but in the long-term individuals are locked into a pattern of response with no escape. This reflects the lives of Jayden and Susan; though the attacks have many negative consequences, they began within very difficult circumstances and had an overall positive impact. Their persistence relates to lack of escape (other behavioural strategies) and the behaviour continues due to the short-term positive consequences.

Further illustrations of attack reduction, contingent on the value and context of positive consequences, have been reported in Susan’s and Jayden’s lives. In the time between her first marriage ending, meeting her new husband and having the TIA, she reported very infrequent NEAs, a time of reduced stress and negative emotion. The significant increase in NEA frequency occurred after
the TIA when life was particularly stressful and she reported increased emotionality. With regard to Jayden, during his participation in the study his current context was considered. When his partner and child went on holiday for ten days he reported having no attacks and a significant reduction in migraines (functionally similar). It is hypothesised that this was due to a reduction in demands, related anger, and fear. Though it has been suggested that stress is a precipitating/immediate trigger for NEAs (Reuber, 2009) this study offers an explanation of how and why this may be the case.

**Do the non-epileptic attacks have similar functional qualities for the different individuals?** All three participants’ NEAs appear to serve to reduce intolerable demands/experiences. For Jayden these seem to be external social demands and subsequently intolerable expression of anger. Susan’s NEAs appear to suppress emotional expression which she feared would be punished due to her childhood experiences. Daisy’s NEAs seem to function to reduce practical demands (though these were often due to her own expectations/rules) and subsequently reduce her fatigue and pain. Though avoidance was been suggested by behaviourists as a primary mechanism in NEAD (Devinsky, 1998), MSFA captures subtle differences in functions which would translate to important implications for treatment. Based on the hypotheses presented here, exposing Jayden and Susan to emotions to disprove their beliefs about the negative consequences of expression may be effective, but for Daisy (even in the peak of her NEAs) this would be inappropriate.

As already mentioned both Jayden’s and Susan’s NEAs appear to be a strategy for suppressing emotional expression. The concept of conversion, proposed by psychoanalytic theorists, suggests that physical symptoms arise to alleviate the conflict of unexpressed emotional pain (particularly related to traumatic early experiences) (Breuer & Freud, 1974). This would suggest NEAs are a means of expressing emotions rather than supressing them as hypothesised in the cases of Jayden and Susan. Additionally both were hypothesised to have NEAs in response to here-and-now stresses and resulting difficult emotions, rather than in response to memories of past traumatic/stressful events. Witnessing attacks in the interviews with Susan offers some verification for this hypothesis. Discussing in detail her difficult childhood and her experiences of physical and
sexual abuse Susan did not have any non-epileptic attacks, whereas beginning to discuss current stressful situation, namely her benefits being reviewed, and her daughter injuring herself and needing more support, Susan had an attack on both occasions. Though a nuanced difference, the psychoanalytic proposition centres on and prioritises early experiences of trauma (Breuer & Freud, 1974), and whilst these hypotheses consider the role of trauma in NEAD development, the priority in maintenance combines learning history with current context.

NEAs becoming generalised to occur in response to internally aversive stimuli (thoughts/emotions) are evidence that experiential avoidance is relevant mechanism in NEAD maintenance (at least for Jayden and Susan). This support the findings of Di Maro and colleagues (2014). Particularly for Susan, persisting NEAs appear to occur when she thinks about current stress, without the need for an external trigger. Though Jayden’s NEAs were initially generalised to occur in response to environmental triggers of anger, and he continues to experience such triggers, it appears that due to the learned negative consequences of expressing anger the thought of experiencing any anger now elicits fear and the need to avoid.

Susan’s early reports of dissociation which re-emerged during her first marriage is the most purposeful strategy described in this study. Though this may be interpreted as evidence for the simulation or volitional control of such behaviour, it is hypothesised that describing it in this way provides Susan with a sense of self-control. Portraying that she was able to do something in the horrendous situations suggested that she was not just weak, which is something she fears being perceived as. Importantly, the functions of the participant’s responses in interviews were considered in relation to their learning histories as verbal reporting behaviour would have also been influenced by previous experiences.

In addition to the negative reinforcement of avoidance, the attacks appeared to be positively reinforced in varying degrees by increased care and support (secondary gains). This appeared most important for Jayden, for whom illness behaviour consistently elicited care from his mother in his childhood. It appears that the increased levels of care and support he received following his head
injury (in response to hypothesised seizures and then NEAs) contributed to increasing self-esteem. This supports the hypothesis made by Moore and colleagues (1994) that comparable levels of self-esteem in NEAD patients and in healthy controls may be due to secondary gains enhancing self-esteem. This may also apply to Susan who reported dissatisfaction regarding others, rather than to the attacks or to herself. Susan’s NEAs were positively reinforced by increased concern from family, friends and professionals, and though this reduced as people become used to them, concern increased as NEA frequency increased following the TIA.

The differing NEA semiology in these cases highlights the issue with suggested heterogeneity. It is unclear what is specifically being referring to when NEAD patients are suggested to be a heterogeneous population, though semiology (Gates et al., 1985) and the lack of CSA in some patients (Betts & Boden, 1992a) seem associated with this proposition. Even within these three cases, those whose attacks manifest similarly (Susan and Daisy) are not those who appear to share functional similarities (Susan and Jayden). Though this is not to suggest that treatment is ever erroneously decided based on attack semiology it highlights the advantages of the MSFA methodology to analyse differences.

How do previously suggested risk factors appear to interact to influence the development of NEAD in these individuals? The suggested risk factors relevant to the participants in this study will be discussed. Due to the functional nature of the study, the relevant intra-psychic traits/factors will be considered within the context of events/circumstances they appear to relate to.

Trauma. All of the participants had experienced trauma in their childhoods (experiencing or witnessing violence and/or emotional abuse). The functional analyses however suggest that these experiences are not always crucial/important in the development of NEAD. Whilst Susan’s early experiences directly led to the development of strategies for suppressing emotions (key in the development of NEAD), Daisy’s traumatic early experiences appeared to have a less important role. Though Daisy witnessed significant domestic violence in her childhood, it was her upbringing in a militant, pressured house (with no fear/threat of violence at that time) which led to her
developing rules about success and achieving (key in the development of NEAD). This exemplifies the utility of MSFA in identifying the development of behaviour in relation to suggested key experiences. A functional behavioural approach allows for explanation beyond the presence and absence of experiences. Based on correlational evidence and confirmation of trauma experience(s), Daisy may have been referred for trauma focused therapy which, based on this analysis, would not target the areas key in the development of NEAD.

**Family relationships.** As no participants reported experiencing CSA the role of family dysfunction in direct relation to NEAD, rather than as a mediator (Nash et al., 1993; Salmon et al., 2003), is relevant. All participants appeared to be raised in families with difficult relationships, which appeared in varying levels to be dysfunctional. Significant conflicts were reported in line with evidence found by Moore and colleagues (1994). A lack of emotional warmth also appeared to be relevant (Binzer et al., 2004). Daisy’s family home seemed to be a highly controlled environment, fitting with Salmon and colleagues (2003) finding that control in the family is relevant to the development of NEAD. However, they specifically investigated it as a mediator between CSA and NEAD, which was not reported by Daisy or the other two participants.

Beyond the presence/absence of specific experiences, functionally early experiences were key in limiting the development of adaptive behaviour. In addition to the impact of physical and emotional abuse (punishing behaviour), social learning theory (Bandura, 1971) would propose that, as children, participants would have learned from their parents’ behaviour. Before Susan’s emotional expression was punished she witnessed it being punished in others and it appeared that Daisy learned to ‘keep up appearances’ by observing the positive impact it had on her mother. This may have contributed to her later behaviour of buying the best things for herself and her children (in conjunction with reinforcement from related behaviour and actual purchases).

**Emotion related deficits.** The hypotheses that Susan learnt to suppress difficult emotions from an early age and therefore had limited experience of adaptive emotional expression, could mean she would be assessed to have
alexithymia characteristics (Sifneos, 1973). The MSFA for Susan supports the proposition of alexithymia as a treatment target rather than a trait useful in differential diagnosis (Bewley et al., 2005; Myers et al., 2013) [See Implications for research and practice, pg 140].

**Stress and coping.** Though all participants reported being faced with many stressful life experiences, it appears that their coping strategies (and/or lack of them) were directly related to the development of NEAD. A lack of task-oriented strategies for coping with stress (Myers, Fleming et al., 2013) appeared to result from a lack of adaptive problem solvers as models in participants’ childhoods.

For all participants NEAs appeared to become a coping strategy for stress/dealing with emotions due to the lack of other strategies and the previous reinforcement of similar behaviour (altered state of consciousness) in a similarly stressful context. NEAs as a coping strategy enabled avoidance which was reinforcing due to short-term relief. Research findings that NEAD patients use more avoidance strategies when faced with stress (Bakvis et al., 2011; Frances et al., 1999; Goldstein et al., 2000; Myers, Fleming et al., 2013) perhaps captured behaviours/strategies that were obscured by the focus on NEAs in this study. Considering the hypotheses in this study that NEAs were the primary avoidance strategy, other avoidance strategies may arise should NEAs be targeted. Indeed, other medically unexplained symptoms arose when NEAs ceased following NEAD diagnosis in a study by McKenzie, Oto, Graham and Duncan, (2011). A behavioural explanation would suggest this would occur through the process of stimulus generalisation. This may also explain the finding that attack reduction or cessation is not associated with other positive outcomes (Reuber et al., 2005).

**Study limitations.**

A significant limitation was the difficulty accessing some historical documents/files for triangulation. This was particularly problematic with Susan’s case, the oldest participant. Time, ethical and practical constraints of the study meant that less older documents were able to be sourced, which may have offered information concerning her adolescence, when it seems that her NEAs
began. Additionally, seemingly due to diagnoses/disorders being missed, Susan reported being sent for further and specialist assessments in other localities, and related correspondence to the home trust (where files were accessed) was sometimes lacking. This resulted in reliance on the other two sources of data in hypothesising some aspects of Susan’s NEAD development and maintenance.

The typical limitations of retrospective reporting were reduced through the triangulation of data. However, a particular concern was the influence of previously suggested ideas and professional opinions on participant and relative reporting/recall. Specifically, the episodes of altered consciousness (e.g. syncope, seizure, NEA) were reported to be whatever they had been diagnosed with, and were explained by whatever they had been told they were related to. This influenced participants and relatives reporting of how they were experienced and perceived at the time. Despite this, the intensive interviews focusing on functional relationships between events and all aspects of behaviour enabled thorough analysis which seemed to enable differentiation between retrospective attributions and experiences at the time.

It is also important to consider the limitations of radical behaviourism (which underpins MSFA), and functional analysis as a method. The most common critique is particularly levelled towards early behavioural theories for reducing complex processes into simple interactions (Chomsky, 1959). More relevant is the argument that it is unethical and inappropriate to extrapolate findings from animal studies to human behaviour (Boulding, 1984; Chomsky, 1959). Whilst radical behaviourism considers language and emotion (Skinner, 1974) thus adding complexity its foundation, operant learning, originated from the findings in animal studies. Though the MSFA framework offers more detail than a single functional analysis, it is still a simplification of Skinner’s full account of human behaviour. Acknowledging the limitations of individual reinforcement schedules in describing complex behaviour, Skinner (1966) introduced the concepts of rule-governed behaviour, chaining and shaping. Including these concepts would have enabled a more comprehensive explanation of NEA, as a seemingly automatic complex behaviour. However, the need for a framework with clinical utility necessitates a more parsimonious application of the theory.
Implications for research and practice. The terms ‘behavioural’ and ‘functional’ seem to have become erroneously associated with the proposition of malingering (Stone et al., 2002). The understanding of NEAD presented here in behavioural and functional terms is not suggesting a conscious or manipulative process, but that the development and maintenance of NEAD can be understood using established behavioural psychological principles. Addressing the limitations of current theories (Baslet, 2011; Brown 2004, 2006; Brown et al., 2011), a functional approach is able to, explain why specifically NEAD develops and produce testable and specific hypotheses/treatment targets. Additionally it offers a more concrete application of behavioural theory, moving beyond social learning theory (Bandura, 1971; Hopkins, 1989; Ramani et al., 1980) and widening the concept of symptom modelling beyond epilepsy (Bautista et al., 2008).

It is suggested that future research should seek to verify hypotheses made using the MSFA framework. A similar study where hypotheses are generated suggesting appropriate explicit treatment targets, followed by such treatment with outcomes assessed is recommended. Due to ethical concerns regarding the professional support network for this populations, it is advised that such a study be located within a current specialist NEAD/functional neurology psychology or psychotherapy service.

Should the MSFA framework prove useful for directing appropriate treatment and further similar hypotheses are found within these studies, further research should assess whether the similarities in NEAD development apply to wider samples of NEAD patients. Template analysis (King, 1998) may be an appropriate method for this once a pattern has been established. Due to the proposition in this study that previously suggested risk factors are both non-specific and common place a template would need to be based on identified mechanisms. Template analysis enables the identification of convergence/divergence with a proposed template (King, 1998), in this case the pathways and functions of NEAD. If a consistent pattern (or a few consistent patterns) are found this may enable the application of less intensive functional analysis in clinical practice in order to ideographically assess and treat NEAD.
This would look similar to the intervention study by Rusch et al. (2001) whereby patients were grouped and treated according to their symptom patterns.

Comparing the three case conceptualisations to each other and to existing literature, it appears Daisy is a less typical NEAD patient. This in itself is interesting for future research, though it is important to consider that Daisy’s NEAD has improved and therefore individuals like her may be less likely to enter treatment and meet the attention of researchers in the field. It is also important to consider the limitations of the risk factor research (earlier detailed) and that psychiatric rather than neurological comparisons groups may be more appropriate (Binzer et al., 2004; Bodde et al., 2011; Kabakoff, 1990; Krawetz, 2001; Rind et al., 1998), and may offer different insights into understanding the population.

This study demonstrates proof of concept for MSFA and functional analysis as an assessment tool with this population. In the study by Mappin et al. (2014) feedback on the case conceptualisations provided evidence that MSFA was acceptable to participants as an assessment and formulation framework. Unfortunately, due to the requirements of the recruiting service the individual formulations were not shared with the participants, and so it was not possible to ascertain this within the current study. It is unknown whether Brown’s integrative cognitive model has been used as a method to communicate explanations of NEAD to patients (Brown, 2004, 2006; Brown et al., 2011). It is proposed that an explanation based on a developmental functional analysis, may be more easily understood. It is however important that experienced clinicians deliver case conceptualisations particularly as a parsimonious behavioural explanation may increase the risk of offence due to misinterpretation (Stone et al., 2002).

Though Brown’s ICM (Brown, 2006) proposed targeting avoidance of feared situations as one of many treatment targets, the findings of this study suggest that avoidance is extended beyond situations. In addition to experiential avoidance (avoidance of introspective experiences) being hypothesised as the key mechanism in NEAD maintenance for Jayden and Susan, it has been found in significant levels in NEAD patients (Di Maro et al., 2014). Should the
previously suggested research identify experiential avoidance as a common mechanism, formal therapies which target this should be tested with this population. Acceptance and Commitment Therapy (ACT; Hayes et al., 1999) is a third-wave cognitive behavioural approach targeting common underpinning mechanisms/processes in psychopathology (one of which being experiential avoidance) (Hayes et al., 2006; Muto & Mitamura, 2011). Alternatively, Intensive Short-Term Dynamic Psychotherapy (ISTDP; Davanloo, 1992, 2005) has a promising evidence base for treating somatisation (Abbass, 2009). ISTDP, underpinned by attachment and psychoanalytic theories, proposes that psychological symptoms/distress, develop from attempts to avoid emotions which are experienced as painful/distressing (due to being punished or invalidated by early attachment figures) (Davanloo, 1980). The psychological presenting symptoms (in this case NEAs) are suggested to be a compromise between expressing the painful emotions and suppressing them using defences; the symptoms represent the channelling of anxiety due to the conflict between expression and defence (Davanloo, 2005). In therapy clients are encouraged and supported to express the emotions which will then reduce the need for defences, the conflict and the resulting symptoms (Davanloo, 1992, 2005). This would target the experiential avoidance (avoidance of emotions) hypothesised to underpin Jayden and Susan’s NEAs. As part of the research process Susan was signposted to a psychotherapist who uses ISTDP for somatisation. Susan expressed an interest in engaging in therapy, the developing hypothesis in conjunction with locally available therapeutic provision indicated that ISTDP might be most helpful. Although this does not verify the hypothesis directly, in the feedback meeting Susan reported that therapy was going well commenting that she was “avoiding her emotions less”.

**Conclusion.** NEAD has been difficult to adequately understand using structural correlational research and models developed from such evidence. Using a functional approach to understand how correlated factors may relate to the process of NEAD development was indicated. Using MSFA to understand the functional development of NEAD in three adult’s histories has produced explanations based on behavioural conditioning processes. This has proposed NEAD as an operant behaviour developing from limited behavioural repertoires.
and altered states of consciousness with positive consequences. Future research to develop and verify hypotheses, and consider wider similarities and differences has important implications for treatment development.

**Critical reflections.**

*The experience of doing the research (my MSFA).* Though this will be presented narratively as opposed to in table form, this section details the self-developed MSFA case conceptualisation of my learning experiences during this thesis project. Autoethnography is a narrative analysis pertaining to oneself as a researcher-practitioner as related to a particular phenomenon (McIlveen, 2008). Many analytic methods and corresponding theoretical positions have been used within autoethnography (Ellis, Adams & Bochner, 2010), behavioural functional analysis may be particularly useful as a means of illustrating specific moments that have significantly impacted the research.

I came to consider this project when my initial project was foreseen to be at high risk of recruitment difficulties. I have never been a particularly indecisive person but I do take time to consider important decisions. Decisions relating to future success (influenced by my parents rewarding success) were important to me, ever since I decided not to go to drama school, opting for a more definite career path in psychology. Mirroring the behaviour I used to make that decision, I sought advice from perceived experts, then it was my parents and tutors, and with this decision it was my research supervisors. This could be interpreted as an adaptive strategy or as avoidance, delaying making a difficult decision, which in the end was a decision I had to make because no-one would make it for me.

Upon taking on the project I felt less anxious about its success than I had done the previous project, which reinforced to me that I had made the right decision. A reduction in anxiety within the context of a highly anxious environment (the doctoral course and my cohort), meant that the decision to stick with this project was a behaviour which was strongly negatively reinforced. However, when my prepared research proposal failed and I was advised by one marker not to continue with the project my anxiety increased again (out of fear of not being successful). Once again I attempted to defer to experts as a strategy to reduce my anxiety. In the short-term this strategy indeed reduced my anxiety, however,
it was around this time I began to recognise that my behaviour was being shaped by my research supervisors. They did not respond how I had previously learned ‘experts’ would, they didn’t tell me what to do. I had to make the decision, as not making a decision was not an option to prevent feared failure. Having already admitted failure by leaving behind a project I had worked hard on, changing again was something I was less likely to do (as it would be further evidence of me being a failure). Importantly I also considered that this project was feasible and something I was particularly interested in, and sticking with it was a behaviour that had been reinforced. I stuck with the project.

Within the data collection and analysis I also avoided making difficult decisions, most notably, the initial hypotheses regarding attack onset. Based on my previous experience hypothesising early experiences, emotions and responses to difficult situations I felt comfortable with most of the analysis. It felt novel for me to be hypothesising how NEAs began. I found myself falling into black and white thinking – trying to work out whether the altered states of consciousness were “organic or not”. Though I was not considering any behaviour as simulated or volitional this difficulty enabled me to empathically understand how NEAs and other behaviour without an identified organic cause could lead to confusion, mismanagement, and avoidance by professionals. Though I vehemently disagree with the preponderant concept of mind/body dichotomy I found myself thinking in such terms which was an uncomfortable realisation. I worked through this in supervision and by relying on the behavioural theory, viewing all behaviour as respondent or operant. Understanding the underpinning origin of a behaviour is less important that understanding its context and consequences in terms of developing hypotheses. If my aim had been to try and offer an explanation for Daisy’s one-off stroke/functional hemiparesis episode for example, the origins may have been more important, but in the persisting NEAs there is much more information available to help understand it.

I developed increasing confidence in the hypotheses that were developing until it appeared that Jayden continues to have two types of ‘attacks’. The hypothesis generated thus far suggested that initial post-head injury seizures had resolved naturally over time and were replaced with NEAs due to their overall positive consequences. Further questioning regarding the two types of attacks led to a
new hypothesis that Jayden developed epilepsy as a result of his head injury and as these have been treated they have reduced in frequency but NEAs were emitted in similar context due to the positive consequences of the seizures.

Completing the file review after the hypothesis was developed I ‘deferred to an expert’ once again. However, the Consultant Neurologists clinical diagnosis/opinion was that Jayden had never had organic seizures and did not have current co-morbid epilepsy and NEAD. This initially made me question my hypothesis, influenced by my strategy of deferring to ‘experts’, this being an actual expert in the field. In a time of anxiety my black and white thinking re-emerged, myself versus a Consultant Neurologist, he must be right and I must be wrong. Examining all of the data I had, I became even more confused and anxious as the expert’s diagnosis/opinion didn’t make sense. I had to re-evaluate my expectation of ‘experts’ in light of this, the thorough clinical interviews and application of behavioural theory has been positively reinforced as it was praised by my supervisors and it offered a complete explanation. I considered the data I had gathered, compared to the data the Consultant Neurologist would have had available to him at the time (and that v-EEG monitoring had never been used to rule out current epileptic seizures) and came to the conclusion that my hypothesis made the most sense. I believe this incident has significantly impacted my expectations of ‘experts’ in the future as well as further strengthening my belief in the explanatory utility of behavioural theory. Though in this capacity I am not responsible for the participants’ treatment the Consultant Neurologist will view the results of the study. However, as a result of this learning experience, I have behaved differently on placement, using thorough functional analyses as evidence to challenge psychiatric opinions.

Taking the nearly completed research to a formative viva panel I was again faced with criticism of the research. Contextually this was at a similar time to growing increasingly angry towards psychiatry and the dominant medical model of explaining (describing) distress on my clinical placement. The criticism at the viva panel was aimed at the MSFA method, suggesting it to be no different to formulation in clinical practice. I felt angry for similar reasons, why particular methods/views are privileged and held in higher regard than others but also
angry that the rigorous research process was being underestimated and my hard work challenged. I had to reflect on my tendency to engage in ‘black and white’ thinking in considering how angry I felt and subsequently how to deal with this criticism. Whilst I felt that MSFA was valuable, rigorous and worthy as a research method, I had to consider that others may see things on a spectrum. They may not have said it was “not a research method” but this was how I interpreted the criticism, because I tend to think of things as either all good or all bad. Recognising this I was able to reflect that strengthening my argument and showing more of why this method was useful, could serve to abate such concerns.

**Ethical issues and further reflections.** Further to the decisions made within my MSFA, additional issues arose during the process for which I have reflected on offering further insight into the project.

Not being able to verify/check out the case conceptualisations with the participants was particularly frustrating for me. As a Trainee Clinical Psychologist, for the most part we learn to be collaborative and additionally that sharing formulations can be interventive in themselves (Johnstone & Dallos, 2014). I wanted to share them in hope that they would have a therapeutic effect but also because I felt uncomfortable not sharing them. It felt as though I was deeming myself the ‘expert’ of their experiences by taking information and analysing it to produce an explanation, or I feared that would be how I would be perceived. Although I have not come to accept myself as the ‘expert’, through the rigorous process of analysis I have become more comfortable in accepting the case conceptualisations as good explanations of the participants’ NEAD development. Utilising multiple data sources and behavioural theory as an additional method of triangulation I now understand the case conceptualisations as a product of the data collection and analysis which may serve to improve understanding of NEAD development.

Upon recruitment to the study none of the participants were engaged in psychological therapies. My interest in psychological therapy for medically unexplained symptoms led to a need for me to balance my longer developed role as a clinician, with my newer role as a researcher. Within the interviews I
asked participants about previous psychological support. Jayden reported currently being on a waiting list for psychology (having been seen in the same service previously) and Susan said that her Consultant Neurologist had referred her to psychology. Daisy had accessed psychology previously but felt she was managing her symptoms and NEAs well, echoed by her Consultant. Finding this out as part of the research reduced my anxieties but with knowledge of the lack of specific psychological support/treatment for NEAD I still experienced some sense of helplessness in the situation. All of the participants reflected that they found the interviews cathartic, and that it was helpful to have a non-judgemental listener. Daisy also commented that it had helped her make sense of some things in the past that may have influenced her behaviour (without the case conceptualisation being fed back).

Undertaking the relative interviews, Jayden’s mother asked for Jayden to be present for her interview. Though it is not explicitly stated (in the protocol) that relative interviews will take place alone, it is intended to be a supplementary independent source of data for triangulation. As it appeared that Jayden’s mother would not be comfortable being interviewed alone, and may have even refused, the decision was made to have Jayden present. Though this may have compromised the supplementary data it conspired to actually produce more information, particularly through their discussion of the context and experiences around the time of Jayden’s father’s death. However, given that Krawetz et al. (2001) found clear differences in reports of family dysfunction given by NEAD patients and their family members separately, Jayden’s mother may have offered different and potentially useful information if she were interviewed alone. An additional concern is that it may have elicited emotions that they are not used to sharing with each other, and it was therefore a potential ethical concern. Though I made the decision at the time and it worked out without causing problems, it led me to consider more thoroughly such decisions in the future.

All of the participants commented on how much they trusted and valued their Consultant Neurologist (the study local collaborator solely responsible for disseminating information sheets). This raised concerns that perhaps they had all been inadvertently coerced into participating due to their reliance on and trust in him. In an attempt to remedy this I made a concerted and perhaps
exaggerated effort to explain that participation was voluntary and that their current support would not be affected if they chose not to take part. I also regularly reminded participants of their right to withdraw. Upon producing a report for an NHS research and development department however, I was asked to report how many prospective participants had been approached. Eight people had been provided with information sheets and only three made contact with me and went on to participate. Though those who did participate may have had a learning history and circumstances leading to them compliance, I would not necessarily suggest this based on the data I had gathered, and this information offered evidence that the majority of those approached did not simply comply.

Though I had a read knowledge that NEAD was widely poorly understood and that diagnostic processes continue to vary, I was nonetheless surprised by the level of variation and professional opinion within the life histories of the participants. This was particularly concerning where medication had been used and seemingly pejorative reports/conclusions were in circulation (including copies being sent to the participants/patients). Additionally, and as reflected in my narrative MSFA, I was concerned that the case conceptualisations disagreed with some key professional opinions. Exploring this I discovered that clinical diagnosis (and not using the best researched methods for diagnosis) is much more common than people would think and perhaps would be lead to believe.

Finally, reflecting on this research process and this study I have found it an experience that has positively reinforced my research behaviour. It is therefore likely I will use this methodology within future research. As a research method it seems that it could be particularly useful to me as a practicing clinical psychologist, with more scope to test the developed hypotheses within treatment.
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Appendices
Appendix A: Ethical Approval.

School of Psychology Research Ethics Committee (SOPREC) correspondence (in order of most recent approval)

notification of non-substantial (minor amendment)

Dear Jenna

This is to confirm that your changes have been approved.

Kind regards

SOPREC

F.A.O SOPREC Chair,

Due to difficulties finding rooms for interviews at the NHS hospitals, a minor amendment has been made to include other sites for interviews.

This has been agreed to be a non-substantial amendment based on REC guidance which includes "inclusion of new sites and investigators in studies other than CTIMPs" as

F.A.O SOPREC Chair,

Due to difficulties finding rooms for interviews at the NHS hospitals, a minor amendment has been made to include other sites for interviews.

This has been agreed to be a non-substantial amendment based on REC guidance which includes "inclusion of new sites and investigators in studies other than CTIMPs" as one of the examples of non-substantial amendments. You can find this guidance here: [https://www.hra.nhs.uk/resources/after-you-applied/amendments/substantial-and-non-substantial-amendments/](https://www.hra.nhs.uk/resources/after-you-applied/amendments/substantial-and-non-substantial-amendments/)

I have attached the documents which were amended as a result, the changes are highlighted. The consent forms have been amended to refer to correct versions of information sheets.

Please could you acknowledge that this minor amendment has been logged for the purposes of a clear audit trail.

Thanks in advance,

Jenna

Jenna Brough
Trainee Clinical Psychologist
Trainee Doctorate in Clinical Psychology
For information.

The current versions of the participant information sheet and consent forms are attached. These include administrative updates - for documents were updated so instructions to contact the chief investigator are consistent throughout the document (as per previous amendments to protocol version 1.2 and documents). This does not change the protocol or research process in any way.

Thanks,
Jenna Brough

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PSY131429 - Jenna Brough - amendment for chair’s action

SUBJECT: Jenna Brough (13451652)

To: Jenna Brough (13451652)

Dear Jenna,

This is to confirm that we are happy with the change.

Regards

SOFFEC

School of Psychology Research Ethics Committee

SOFREC
College of Social Science
University of Lincoln, Bayford Building, Lincoln, Lincolnshire, LN6 7TS
Email: soffec@lincoln.ac.uk

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Jenna Brough (13451652)

To: Jenna Brough (13451652)

Good evening,

Can I just confirm that I don’t need approval for this most recent change (see below), as it is an administrative change.

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Jenna Brough (13451652)

To: Jenna Brough (13451652)

Good evening,

Can I just confirm that I don’t need approval for this most recent change (see below), as it is an administrative change.

Thanks in advance,
Jenna Brough

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Jenna Brough (13451652)

To: Jenna Brough (13451652)

FAO Patrick Bourne - amendments to project.

Hello again,

Unfortunately, the administrative support (outlined in the previous amendment email) to pass over contact details for interested patients is no longer available. Therefore, my contact details will be included on participant information sheets instead and interested patients will be contacting me directly by email or phone. This means the information sheet and consent form has been updated (attached).

The protocol has also been updated to reflect this minor change (attached) and include the new versions of the information sheet and consent form. With regard to my previous email - the previous amendments regarding the final session and sharing of the explanations remain the same pending approval from you and the NRES REC.

Thanks,
Jenna
PSY131429 - Jenna Brough - amendments for chair's action

Subject

Hi Jenna

This is to confirm that your changes have been approved.

Regards

SOPME

Jenna Brough (13451652)

Reply to: Jenna Brough (13451652)

Following discussions with the local collaborator who is supporting the recruitment some amendments have been made to the project. Please note these have also been submitted to the NRES and we will not be moving forward with the project until approval has been received from you and the NRES.

For your information the changes are:

- to include a medical secretary as a contact point for potential participants (to securely email me a contact number for potential participants when they decide they are interested in the neurologist only works for the last 1 day a week and I'm being responsible for this would significantly slow down the process).
- a change to the final section - participants will no longer receive a paper copy of the explanation of their attacks, and it will not be shared with professionals. This was decided to ensure the optimisation of the experience as research not clinical work. More importantly, if they are not yet accepting of a psychological explanation, or if they disagree with it, it may put them off accessing psychology in the future (something which they may benefit from), with regard to not sharing with professionals it is possible that professionals (e.g. psychologists) who participate in the future may not work within a behavioural model and therefore the explanation may not be utilized by or useful for them. Also, the waiting lists mean that it may be a long time before they access psychology and lots of things may have changed by then. Instead the final section will be a discussion of all the information gathered relevant to the explanation which the participant will be encouraged to offer their comments on. Participants and interviewees (relatives/ professionals) will still be offered an executive summary of the results of the study.

These changes involve edits to the protocol, information sheets, consent forms, and interview guide – all of which are attached to this email. The changes have been highlighted and are explained further in the attached Amendment Form which has also been submitted to the NRES.

Thank you in advance,

Jenna Brough

Trainee Clinical Psychologist

Trainee Electorate in Clinical Psychology
PSY13429 - Jenna Brough - further amendments following REC review

Dear Jenna,

Thank you for your email. There is nothing else required.

Kind regards,

SOPREC

Jenna Brough (13451652)

2014/10/29 15:42

[Image 1]

14.00.121.14 ProvOp.pdf

16.38.02.25.16 ProvOp.pdf

14.00.121.14 ProvOp.pdf

PAC - amendments following REC review

Please see attached documents which include the provisional favourable opinion letter from the REC, a document outlining the amendments made, updated versions of the study documents including consent forms, and the REC database itself.

By reading the favourable opinion letter from the REC and then the document "Response to 12122/7" you will see where clarification by the REC has been requested to. Do I need to send a copy of my confirmed favourable opinion letter when it arrives?

The major differences/changes made to the project/documents since receiving SOPREC ethical approval are as follows:

- Participants’ GP will be informed of their participation in the study via a standard letter - this is explained in the information sheet and is a point on the consent form.
- Clarification around when participants can withdraw data (up to point of analysis) - previously it stated 6 weeks the last meeting with the researcher; however, the last meeting is when the produced developmental account is shared and discussed and the analysis will be completed by this point. Clarifications made to participant information sheet and consent form to make it clear that data can be withdrawn up to two weeks after the last interview with the researcher (ie. an example framework: this is interview 4, not the feedback meeting).
- Interview guide has been developed.
- Inclusion of PACS information for consent.
- Inclusion of REC in "who has reviewed the study" section of information sheets.

Please let me know if there is anything I need to do or if anything is unclear.

Thanks in advance,

Jenna Brough
Trainee Clinical Psychologist

Application for SOPREC - PSY13429 - Jenna Brough - amendments

Dear Jenna,

Thanks for your email. This is to confirm that your ethical approval form has now been approved.

Kind regards

School of Psychology Research Ethics Committee
C.advance
College of Social Science
University of Lincoln, Brayford Pool, Lincoln, LN6 7TS
Email - joprec@lincoln.ac.uk

Jenna Brough (13451652)

2014/10/29 15:42

[Image 2]

Track8_DChiPsych_CHI.png

Downloaded

Save to OneDrive - University of Lincoln

Page 183 of 248
Please word-process this form, handwritten applications will not be accepted

Name of Applicant

Jenna Brough
<table>
<thead>
<tr>
<th>School:</th>
<th>College:</th>
</tr>
</thead>
<tbody>
<tr>
<td>Psychology</td>
<td>Doctorate in Clinical Psychology</td>
</tr>
</tbody>
</table>

| 2 Position in the University  | Post-graduate researcher            |
| 3 Role in relation to this research | Chief Investigator                 |
| 4 Brief statement of main Research Question | How do non-epileptic attacks appear to develop in the histories of a sample of adults diagnosed with non-epileptic attack disorder (NEAD)? |
| 5 Brief Description of Project | Using Multiple Sequential Functional Analysis (MSFA) to identify potential developmental pathways of Non-Epileptic Attack Disorder (NEAD) |

This project is a series of case studies exploring the development of NEAD in between three and six adults. The project focuses on developing an individual psychological explanation of non-epileptic attacks for each participant using Multiple Sequential Functional Analysis (MSFA). MSFA is a form of functional analysis based on behavioural principles and will be used to identify functional relationships between risk factors identified in previous literature. The psychological explanations are developed through gathering data from multiple sources: comprehensive interviews with participants, an additional interview with a relative or professional, and a comprehensive review of relevant medical/social care files. The individual cases will be compared to identify any similarities or distinct features which may contribute to theory development and direct future research. The overall purpose of this project is to develop an understanding of the functional development of NEAD. A full research protocol can be found at the end of this document.

<table>
<thead>
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<th>Approximate Start Date:</th>
<th>Approximate End Date:</th>
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</thead>
<tbody>
<tr>
<td>September 2014</td>
<td>January 2016</td>
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</table>

<table>
<thead>
<tr>
<th>6 Name of Principal Investigator or Supervisor</th>
<th>Jenna Brough</th>
</tr>
</thead>
<tbody>
<tr>
<td>Email address:</td>
<td><a href="mailto:13451652@students.lincoln.ac.uk">13451652@students.lincoln.ac.uk</a></td>
</tr>
<tr>
<td>Telephone:</td>
<td>07540613882</td>
</tr>
</tbody>
</table>
Statement of the ethical issues involved and how they are to be addressed – including a risk assessment of the project based on the vulnerability of participants, the extent to which it is likely to be harmful and whether there will be significant discomfort. (This will normally cover such issues as whether the risks/adverse effects associated with the project have been dealt with and whether the benefits of research outweigh the risks)

The prospective and eligible participants are considered ‘vulnerable’ as they will be accessing NHS services for support with their diagnosis of NEAD, a psychological disorder. They may also have co-morbid psychiatric diagnoses such as anxiety and/or depression.

The following details the ethical issues identified and the plans in place to address them.

Informed Consent

All identified eligible prospective participants will be fully informed of the nature of the research through the detailed information sheet (See Appendix A). All participants will be required to formally, through signature on the consent form (Appendix B), indicate their consent to participating in the research process. Participants will be consenting to: one-to-one interviews, an interview with an agreed relative or professional (who will also need to provide informed consent), and a comprehensive file review. Participants will be encouraged to ask questions if anything is unclear or not explained.

Right to withdraw

It will be emphasised throughout the process that participation is entirely voluntary and that if they decide not to take part or to withdraw their care will not be affected. Participants will be informed (information sheet – Appendix A) of their right to withdraw from the research process at any time without giving a reason. This be also be repeated on the consent form and verbally before each interview. They will also be informed that they are free to choose not to answer any individual questions within the interviews without giving a reason. The will also be made aware that they can withdraw their data (interview recordings and notes made) up to two weeks after their last meeting with the

<table>
<thead>
<tr>
<th>Names of other researchers or student investigators involved</th>
</tr>
</thead>
<tbody>
<tr>
<td>1. Dr Mark Gresswell, University of Lincoln, Co-Course Director, Doctorate in Clinical Psychology.</td>
</tr>
<tr>
<td>2. Dr Nima Moghaddam, University of Lincoln, Research Tutor.</td>
</tr>
<tr>
<td>3. Dr Dave Dawson, University of Lincoln, Research Tutor.</td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>Location(s) at which project is to be carried out</th>
</tr>
</thead>
</table>
Recruitment will take place in one or more NHS clinical sites within the region. Interviews will take place on NHS clinical sites and file reviews will take place where the files are stored. Sites will be confirmed once proof of university ethical approval is obtained prior to NHS ethical approval being sought.

<table>
<thead>
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</thead>
</table>
Recruitment will take place in one or more NHS clinical sites within the region. Interviews will take place on NHS clinical sites and file reviews will take place where the files are stored. Sites will be confirmed once proof of university ethical approval is obtained prior to NHS ethical approval being sought.
chief investigator. If data is withdrawn it will be destroyed and not included in
the study.

Confidentiality and data protection.

Before consent is obtained and interviews begin, participants (and
relatives/professionals) will be made aware of the confidentiality limits. The
chief investigator will make it clear that any identified/reported concern about
the participant’s safety or the safety of others will be taken forward following
the relevant trust and university policies. Any decision to take forward a
concern will be discussed with the participant unless it is felt that this would
increase the concern for their safety of the safety of others.

Interview audio recordings will be transferred onto a secure laptop and erased
from the Dictaphone at the first available opportunity. All paper records
including notes from interviews and file reviews will be anonymous.
Participants (including relatives/professionals) will be assigned a pseudonym
at the beginning of their participation for differentiating and storing data, and
for referring to interview excerpts in the final thesis and journal submission.
Any need to transport data between secure laptops will utilise an encrypted
memory stick.

Consent forms containing personal data will be stored securely in a locked
cabinet in a locked office at the University of Lincoln. All data at all stages will
be treated with strict confidence and will be accessible to the chief
investigator, research supervisors, administrators and auditors/regulatory
bodies from the NHS and the University of Lincoln. All data related to the
research will be stored for seven years after the completion of the study in
accordance with university regulations and following this will be destroyed
securely. Identifiable data will be destroyed securely three months following
completion of the study.

Addressing concerns/questions

The researcher will offer prospective participants the chance to ask questions
before they consent to take part and at the beginning and end of each
interview. Contact details for the research supervisors and SOPREC will be
detailed on the participant information sheet (Appendix A) if they have any
concerns or questions they feel they cannot approach the researcher with.

Protection of research participants

The focus of the study is the development of NEAD in adults with this
diagnosis. The interviews will include questions about all aspects of the
participants’ lives which may lead to discussions about
negative/upsetting/distressing events. The participant may also be exposed to
new potentially distressing information (resulting from the data gathered from
other sources) when the MSFA is discussed with them. The potential for
distress will be discussed before consent is obtained, and before and after
each interview session. The chief investigator has considerable experience of
interviewing vulnerable adults as part of their profession and also containing/managing any resulting distress. It is hoped that the chief investigator will be able to contain and manage moderate levels of distress. If any participant asks for or requires further support they will be referred to their current care network (Clinical Psychologist/GP) as appropriate. Consent to discuss any reported or identified distress with a current professional will be sought.

The research supervisors, Dr Mark Gresswell, Dr Nima Moghaddan and Dr David Dawson (and a yet to be identified field supervisor) and SOPREC will be available to contact for any concerns related to the chief investigator or any element of the research.

Debriefing of research participants

Participants will be involved in the development of the individual psychological explanation of the development of their non-epileptic attacks and will meet with the chief investigator once it is developed to give their opinion and feedback. Participants will be offered their finalised MSFA case conceptualisation and an executive summary of the study following its completion.

Inconvenience allowance

Recognising that taking part in the study will involve a considerable time commitment from participants, they be afforded an inconvenience allowance of a £5 high street gift voucher per hour of their time for the interviews. As the interviews may take up to 7 hours the participants will be paid up to £35 in gift vouchers. Participants who withdraw during the process will be paid for their time so far (£5 per hour). Participants will be made aware of this through the participant information sheet (Appendix A).

Many of the same (and no additional) ethical issues have been identified as applying to the relatives and/or professionals who will be interviewed. Separate information and consent forms have been produced (see Appendices C and D).

For more information please see attached protocol and appendices.
26 November 2015
Miss Jenna L Brough
45 Cambridge Avenue
Bottesford
Scunthorpe
DN16 3PH

Dear Miss Brough

Study title: Using Multiple Sequential Functional Analysis (MSFA) to identify potential developmental pathways of Non-Epileptic Attack Disorder (NEAD)

REC reference: 14/WA/1214
Amendment number: 02
Amendment date: 08 June 2015
IRAS project ID: 155459

Thank you for your letter of 08 June 2015, notifying the Committee of the above amendment.

You advised that due to difficulties finding rooms for interviews at the NHS hospitals, a minor amendment has been made to include other sites for interviews.

Confirmation was provided by you that his has been agreed with the sponsor to be a non-substantial amendment based on REC guidance which includes "inclusion of new sites and investigators in studies other than CTIMPs" as one of the examples of non-substantial amendments.

Participant documentation was amended as a result for clarification. The consent forms have been amended to refer to correct versions of information sheets.
As home visits may potentially be involved you confirmed adherence to the Trust Lone Worker Policy. Further assurance was provided that the Neurologist will also be advised of your whereabouts should a home visit be undertaken and contact will also be made at the end of the visit day.

Further confirmation was provided by you that participant initials only will be provided to the Neurologist to enable location to be found if no contact has been made at the end of the home visit.

The Committee does not consider this to be a “substantial amendment” as defined in the Standard Operating Procedures for Research Ethics Committees. The amendment does not therefore require an ethical opinion from the Committee and may be implemented immediately, provided that it does not affect the approval for the research given by the R&D office for the relevant NHS care organisation.

Documents received

The documents received were as follows:

<table>
<thead>
<tr>
<th>Document</th>
<th>Version</th>
<th>Date</th>
</tr>
</thead>
<tbody>
<tr>
<td>Notice of Minor Amendment</td>
<td>02</td>
<td>08 June 2015</td>
</tr>
<tr>
<td>Participant information sheet (PIS)</td>
<td>2.4</td>
<td>06 June 2015</td>
</tr>
<tr>
<td>Participant consent form</td>
<td>2.4</td>
<td>06 June 2015</td>
</tr>
<tr>
<td>Participant information sheet (PIS) [Relative/Professional]</td>
<td>2.2</td>
<td>06 June 2015</td>
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<tr>
<td>Participant consent form [Relative/Professional]</td>
<td>2.2</td>
<td>06 June 2015</td>
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</table>

Statement of compliance

The Committee is constituted in accordance with the Governance Arrangements for Research Ethics Committees and complies fully with the Standard Operating Procedures for Research Ethics Committees in the UK.

14/WA/1214: Please quote this number on all correspondence

Yours sincerely

Mrs Tracy Biggs
Research Ethics Committee Manager

E-mail: tracy.biggs@wales.nhs.uk
Thank you for your email notifying the Committee of the above amendment. The minor amendments to the protocol were:

i) To remove appendices from the protocol (due to issues with ensuring version control of appended documents within the document if the protocol itself does not need to change).

ii) To change who participants contact if interested - instead of contacting Neurology admin, participants will be contacting CI directly by email or phone.

The Committee does not consider this to be a “substantial amendment” as defined in the Standard Operating Procedures for Research Ethics Committees. The amendment does not therefore require an ethical opinion from the Committee and may be implemented immediately, provided that it does not affect the approval for the research given by the R&D office for the relevant NHS care organisation.
Documents received

The documents received were as follows:

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<tr>
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<th>Version</th>
<th>Date</th>
</tr>
</thead>
<tbody>
<tr>
<td>Notice of Minor Amendment</td>
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<td>05 June 2015</td>
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<td>Research protocol or project proposal</td>
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<td>29 January 2015</td>
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Statement of compliance

The Committee is constituted in accordance with the Governance Arrangements for Research Ethics Committees and complies fully with the Standard Operating Procedures for Research Ethics Committees in the UK.

14/WA/1214: Please quote this number on all correspondence

Yours sincerely

Mrs Tracy Biggs
Research Ethics Committee Manager

E-mail: tracy.biggs@wales.nhs.uk
27 February 2015

Miss Jenna L Brough
45 Cambridge Avenue
Bottesford
Scunthorpe
DN16 3PH

Dear Miss Brough

Study title: Using Multiple Sequential Functional Analysis (MSFA) to identify potential developmental pathways of Non-Epileptic Attack Disorder (NEAD)

REC reference: 14/WA/1214
Amendment number: 1
Amendment date: 27 January 2015
IRAS project ID: 155459

The above amendment was reviewed at the meeting of the Sub-Committee held on 26 February 2015.

Ethical opinion
The members of the Committee taking part in the review gave a favourable ethical opinion of the amendment on the basis described in the notice of amendment form and supporting documentation.

Approved documents
The documents reviewed and approved at the meeting were:

<table>
<thead>
<tr>
<th>Document</th>
<th>Version</th>
<th>Date</th>
</tr>
</thead>
<tbody>
<tr>
<td>Interview schedules or topic guides for participants</td>
<td>1.1</td>
<td>26 January 2015</td>
</tr>
<tr>
<td>Notice of Substantial Amendment (non-CTIMP)</td>
<td>1</td>
<td>27 January 2015</td>
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<tr>
<td>Participant consent form</td>
<td>2.3</td>
<td>25 February 2015</td>
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<tr>
<td>Participant consent form [Relative/Professional]</td>
<td>2.1</td>
<td>26 January 2015</td>
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<tr>
<td>Participant information sheet (PIS) [Relative/Professional]</td>
<td>2.1</td>
<td>26 January 2015</td>
</tr>
<tr>
<td>Participant information sheet (PIS)</td>
<td>2.3</td>
<td>25 February 2015</td>
</tr>
</tbody>
</table>
Membership of the Committee
The members of the Committee who took part in the review are listed on the attached sheet.

R&D approval
All investigators and research collaborators in the NHS should notify the R&D office for the relevant NHS care organisation of this amendment and check whether it affects R&D approval of the research.

Statement of compliance
The Committee is constituted in accordance with the Governance Arrangements for Research Ethics Committees and complies fully with the Standard Operating Procedures for Research Ethics Committees in the UK.

We are pleased to welcome researchers and R & D staff at our NRES committee members’ training days – see details at http://www.hra.nhs.uk/hra-training/

14/WA/1214: Please quote this number on all correspondence

Yours sincerely

E-mail: tracy.biggs@wales.nhs.uk

Enclosures: Names and professions of members who took part in the review
Copy to: NNHS R&D contact - Helen Ayre, United Lincolnshire Hospitals NHS Trust
Sponsor contact - Professor Sara Owen

Wales REC 4
Attendance at Sub-Committee of the REC meeting on 26 February 2015

Committee Members:

<table>
<thead>
<tr>
<th>Name</th>
<th>Profession</th>
<th>Present</th>
<th>Notes</th>
</tr>
</thead>
<tbody>
<tr>
<td>Professor Alex Carson - Chair</td>
<td>Retired</td>
<td>Yes</td>
<td></td>
</tr>
<tr>
<td>Dr Kath Clarke</td>
<td>Deputy Associate Chief of Staff, Nursing</td>
<td>Yes</td>
<td></td>
</tr>
<tr>
<td>Mr Philip Richards</td>
<td>Associate Specialist - Surgery</td>
<td>Yes</td>
<td></td>
</tr>
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</table>

Also in attendance:

<table>
<thead>
<tr>
<th>Name</th>
<th>Position (or reason for attending)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Mrs Tracy Biggs</td>
<td>Research Ethics Committee Manager</td>
</tr>
</tbody>
</table>
05 December 2014

Miss Jenna L Brough
45 Cambridge Avenue
Bottesford
Scunthorpe
DN16 3PH

Dear Miss Brough

Study title: Using Multiple Sequential Functional Analysis (MSFA) to identify potential developmental pathways of Non-Epileptic Attack Disorder (NEAD)

REC reference: 14/WA/1214
IRAS project ID: 155459

Thank you for your letter received 26 November 2014, responding to the Committee’s request for further information on the above research and submitting revised documentation.

The further information was considered by a Sub-Committee of the REC at a meeting held on 03 December 2014. A list of the Sub-Committee members is attached.

We plan to publish your research summary wording for the above study on the HRA website, together with your contact details. Publication will be no earlier than three months from the date of this opinion letter. Should you wish to provide a substitute contact point, require further information, or wish to make a request to postpone publication, please contact the REC Manager, Mrs Tracy Biggs, Tracy.Biggs@Wales.nhs.uk.

Confirmation of ethical opinion
On behalf of the Committee, I am pleased to confirm a favourable ethical opinion for the above research on the basis described in the application form, protocol and supporting documentation as revised, subject to the conditions specified below.

Conditions of the favourable opinion
The favourable opinion is subject to the following conditions being met prior to the start of the study.
Management permission or approval must be obtained from each host organisation prior to the start of the study at the site concerned.

Management permission ("R&D approval") should be sought from all NHS organisations involved in the study in accordance with NHS research governance arrangements.

Guidance on applying for NHS permission for research is available in the Integrated Research Application System or at http://www.rdforum.nhs.uk.

Where a NHS organisation’s role in the study is limited to identifying and referring potential participants to research sites ("participant identification centre"), guidance should be sought from the R&D office on the information it requires to give permission for this activity.

For non-NHS sites, site management permission should be obtained in accordance with the procedures of the relevant host organisation.

Sponsors are not required to notify the Committee of approvals from host organisations.

Registration of Clinical Trials

All clinical trials (defined as the first four categories on the IRAS filter page) must be registered on a publicly accessible database within 6 weeks of recruitment of the first participant (for medical device studies, within the timeline determined by the current registration and publication trees).

There is no requirement to separately notify the REC but you should do so at the earliest opportunity e.g when submitting an amendment. We will audit the registration details as part of the annual progress reporting process.

To ensure transparency in research, we strongly recommend that all research is registered but for non clinical trials this is not currently mandatory.

If a sponsor wishes to contest the need for registration they should contact Catherine Blewett (catherineblewett@nhs.net), the HRA does not, however, expect exceptions to be made. Guidance on where to register is provided within IRAS.

It is the responsibility of the sponsor to ensure that all the conditions are complied with before the start of the study or its initiation at a particular site (as applicable).

Ethical review of research sites

NHS sites

The favourable opinion applies to all NHS sites taking part in the study, subject to management permission being obtained from the NHS/HSC R&D office prior to the start of the study (see "Conditions of the favourable opinion" below).
Non-NHS sites

Approved documents
The final list of documents reviewed and approved by the Committee is as follows:

<table>
<thead>
<tr>
<th>Document</th>
<th>Version</th>
<th>Date</th>
</tr>
</thead>
<tbody>
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<td>Covering letter on headed paper [RESPONSE TO REC]</td>
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<tr>
<td>Evidence of Sponsor insurance or indemnity (non NHS Sponsors only) [Sponsor indemnity insurance]</td>
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<td>GP/consultant information sheets or letters [GP letter]</td>
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<tr>
<td>Interview schedules or topic guides for participants [Interview guide]</td>
<td>1.0</td>
<td>19 October 2014</td>
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<td>IRAS Checklist XML [Checklist_24102014]</td>
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<tr>
<td>Participant consent form [consent form]</td>
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<tr>
<td>Participant consent form [relative/professional consent]</td>
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<td>Participant information sheet (PIS) [PIS]</td>
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</tr>
<tr>
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<td></td>
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<tr>
<td>Referee's report or other scientific critique report [academic feedback]</td>
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<td>08 August 2014</td>
</tr>
<tr>
<td>Research protocol or project proposal [Protocol]</td>
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<td>19 October 2014</td>
</tr>
<tr>
<td>Summary CV for Chief Investigator (CI) [CI CV]</td>
<td>1.0</td>
<td>22 October 2014</td>
</tr>
<tr>
<td>Summary CV for supervisor (student research) [Supervisor CV]</td>
<td>1.0</td>
<td>01 October 2014</td>
</tr>
</tbody>
</table>

Statement of compliance
The Committee is constituted in accordance with the Governance Arrangements for Research Ethics Committees and complies fully with the Standard Operating Procedures for Research Ethics Committees in the UK.

After ethical review
Reporting requirements
The attached document “After ethical review – guidance for researchers” gives detailed guidance on reporting requirements for studies with a favourable opinion, including:

- Notifying substantial amendments
- Adding new sites and investigators
- Notification of serious breaches of the protocol
- Progress and safety reports
- Notifying the end of the study
The HRA website also provides guidance on these topics, which is updated in the light of changes in reporting requirements or procedures.

User Feedback
The Health Research Authority is continually striving to provide a high quality service to all applicants and sponsors. You are invited to give your view of the service you have received and the application procedure. If you wish to make your views known please use the feedback form available on the HRA website: http://www.hra.nhs.uk/about-the-hra/governance/quality-assurance/

HRA Training
We are pleased to welcome researchers and R&D staff at our training days – see details at http://www.hra.nhs.uk/hra-training/

14/WA/1214 Please quote this number on all correspondence

With the Committee’s best wishes for the success of this project.

Yours sincerely

Professor Alex Carson
Chair

E-mail: tracy.biggs@wales.nhs.uk

Enclosures: List of names and professions of members who were present at the meeting and those who submitted written comments

“After ethical review – guidance for researchers”

Copy Sponsor contact - Professor Sara Owen

to: Lead NHS R&D Contact - Helen Ayre, United Lincolnshire Hospitals NHS Trust

Wales REC 4

Attendance at Sub-Committee of the REC meeting on 03 December 2014

Committee Members:

<table>
<thead>
<tr>
<th>Name</th>
<th>Profession</th>
<th>Present</th>
<th>Notes</th>
</tr>
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<tr>
<td>Professor Alex Carson - Chair</td>
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<td>Yes</td>
<td></td>
</tr>
<tr>
<td>Dr Kath Clarke</td>
<td>Deputy Associate Chief of Staff, Nursing</td>
<td>Yes</td>
<td></td>
</tr>
</tbody>
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Also in attendance:

<table>
<thead>
<tr>
<th>Name</th>
<th>Position (or reason for attending)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Mrs Tracy Biggs</td>
<td>Research Ethics Committee Manager</td>
</tr>
</tbody>
</table>
12 November 2014

Miss Jenna L. Brough
45 Cambridge Avenue
Bottesford
Scunthorpe
DN16 3PH

Dear Miss Brough

Study Title: Using Multiple Sequential Functional Analysis (MSFA) to identify potential developmental pathways of Non-Epileptic Attack Disorder (NEAD)

REC reference: 14/WA/1214
IRAS project ID: 155459

The Research Ethics Committee reviewed the above application at the meeting held on 05 November 2014. Thank you for being available on the telephone to discuss the application.

Provisional opinion

The Committee would be content to give a favourable ethical opinion of the research, subject to receiving a complete response to the request for further information set out below.

Authority to consider your response and to confirm the Committee’s final opinion has been delegated to a meeting of the Sub committee of the REC.

Further information or clarification required

i. Please provide clarification in respect of what action will be taken if someone suffers an episode.

ii. The response at Question A40 refers to Administration staff at the University of Lincoln being responsible for ensuring the safe storage of personal identifiable data. Please provide full clarification about their role.

iii. Participant Information Sheet should clearly explain why a participant has been chosen. It would be preferable to use the word ‘you’ rather than ‘participant or patient’.

iv. Participant Information sheet and consent form to be revised to include notification that you intend to inform the GP and the option to consent thereto.

v. Participant Information Sheet – Invitation paragraph should indicate that the study is being undertaken as an educational project. ‘Who has reviewed this study?’ Wales REC 4 should be identified as the REC reviewing body.
If you would find it helpful to discuss any of the matters raised above or seek further clarification from a member of the Committee, you are welcome to contact the REC Manager in the first instance.

When submitting a response to the Committee, the requested information should be electronically submitted from IRAS. A step-by-step guide on submitting your response to the REC provisional opinion is available on the HRA website using the following link: http://www.hra.nhs.uk/nhs-research-ethics-committee-rec-submitting-response-provisional-opinion/

Please submit revised documentation where appropriate underlining or otherwise highlighting the changes which have been made and giving revised version numbers and dates. You do not have to make any changes to the REC application form unless you have been specifically requested to do so by the REC.

The Committee will confirm the final ethical opinion within a maximum of 60 days from the date of initial receipt of the application, excluding the time taken by you to respond fully to the above points. A response should be submitted by no later than 12 December 2014.

Summary of the discussion at the meeting

**Care and protection of research participants: respect for potential and enrolled participants’ welfare and dignity**

Clarification surrounding what safeguard is in place in the event of a participant suffering an episode or attack is required.

**Informed consent process and the adequacy and completeness of participant information**

Revision required to explain that the study is being undertaken as an education project and that the GP would be informed if consent if given.

**Suitability of the applicant and supporting staff**

Confirmation was received by you that lone work policies would be adhered to at all times notwithstanding all interviews would take place on NHS premises in rooms with alarms.

Documents reviewed

The documents reviewed at the meeting were:

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<th>Document</th>
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<tr>
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</tr>
<tr>
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<tr>
<td>Summary CV for supervisor (student research) [Supervisor CV]</td>
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</table>
Membership of the Committee

The members of the Committee who were present at the meeting are listed on the attached sheet.

Statement of compliance

The Committee is constituted in accordance with the Governance Arrangements for Research Ethics Committees and complies fully with the Standard Operating Procedures for Research Ethics Committees in the UK.

14/WA/1214 Please quote this number on all correspondence

Yours sincerely

[Signature]

Professor Alex Carson
Chair
E-mail: tracy.biggs@wales.nhs.uk

Enclosures: List of names and professions of members who were present at the meeting and those who submitted written comments.

Copy to: Sponsor contact - Professor Sara Owen
Lead NHS R&D contact - Helen Ayre, United Lincolnshire Hospitals NHS Trust
Wales REC 4  
Attendance at Committee meeting on 05 November 2014

Committee Members:

<table>
<thead>
<tr>
<th>Name</th>
<th>Profession</th>
<th>Present</th>
<th>Notes</th>
</tr>
</thead>
<tbody>
<tr>
<td>Mrs Celia Blomeley</td>
<td>Retired Assistant Headteacher</td>
<td>Yes</td>
<td></td>
</tr>
<tr>
<td>Professor Alex Carson - Chair</td>
<td>Retired</td>
<td>Yes</td>
<td></td>
</tr>
<tr>
<td>Dr Kath Clarke</td>
<td>Deputy Associate Chief of Staff, Nursing</td>
<td>No</td>
<td></td>
</tr>
<tr>
<td>Dr John Clifford</td>
<td>Consultant Psychiatrist</td>
<td>Yes</td>
<td></td>
</tr>
<tr>
<td>Dr John Delieu</td>
<td>Anatomist &amp; DI for HTA Licence</td>
<td>Yes</td>
<td></td>
</tr>
<tr>
<td>Mr John Gittins</td>
<td>Coroner</td>
<td>No</td>
<td></td>
</tr>
<tr>
<td>Mrs Yvonne Harding</td>
<td>Associate Chief of Staff (Nursing) Children &amp; Young People</td>
<td>Yes</td>
<td></td>
</tr>
<tr>
<td>Miss Joy Hickman</td>
<td>Consultant Orthodontist</td>
<td>No</td>
<td></td>
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<tr>
<td>Dr Peter Hobson</td>
<td>Principal Healthcare Scientist (Research)</td>
<td>No</td>
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<tr>
<td>Ms Alison Ledward</td>
<td>Former Midwife/Current Researcher</td>
<td>Yes</td>
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<tr>
<td>Mr Philip Richards</td>
<td>Associate Specialist - Surgery</td>
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<tr>
<td>Dr David Southern</td>
<td>Consultant Anaesthetian</td>
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<td>Ms Eunice Vincent</td>
<td>Retired Nurse/Nurse Lecturer</td>
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<tr>
<td>Dr Anthony White</td>
<td>Consultant Care of the Elderly</td>
<td>Yes</td>
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Also in attendance:

<table>
<thead>
<tr>
<th>Name</th>
<th>Position (or reason for attending)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Mrs Tracy Biggs</td>
<td>Research Ethics Committee Manager</td>
</tr>
</tbody>
</table>
Research and Development departments in NHS trusts (amendments approved or acknowledged by the NHS REC and SOPREC were forwarded to all NHS R&D departments for their records)

Dear Ms Brough,

Re: Using Multiple Sequential Functional Analysis (MSFA) to identify potential developmental pathways of Non Epileptic Attack Disorder (NEAD)

Thank you for submitting the above project for local research governance approval. I am pleased to inform you that your project has been given full approval and you may begin your research at the following site:

- United Lincolnshire Hospitals NHS Trust – Pilgrim Hospital, Boston

The final list of documents reviewed and approved are as follows:

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<th>Document</th>
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</tr>
<tr>
<td>Participant consent form [relative/professional consent]</td>
<td>2.0</td>
<td>23 November 2014</td>
</tr>
</tbody>
</table>

Please notify the R&D department of any future amendments to the approved study and/or documents along with a copy of any Research Ethics Committee correspondence/approval.

Conditions of Approval:
- All researchers involved in Clinical Research must have up to date GCP training – See LCRF Training SOP09 for further details. Available at [http://www.ulh.nhs.uk/for_staff/lincolnshire_crf/documents/sop/SOP09_Training_Record.pdf](http://www.ulh.nhs.uk/for_staff/lincolnshire_crf/documents/sop/SOP09_Training_Record.pdf)
You must ensure that any reports on the progress and/or outcome of your research requested by R&D are produced on time and to an acceptable standard, in accordance with your responsibilities under section 3.6.3 of the Research Governance Framework, 2nd Edition (DOH, 2005). As a minimum, this will include completion of the ULHT R&D annual progress report.

Please note that the Trust audits 10% of ULH non sponsor studies and we anticipate auditing 100% of ULHT sponsored projects approved, on an annual basis.

Please note that should a Suspected Unexpected Serious Adverse Reaction (SUSAR) or complaint arise from this research, the Research & Development department must be informed within 24 hours of identification.

Please Note that should a Serious Adverse Event (SAE) arise from this research, the Research Sponsor must be informed within 24 hours of identification. In the case of ULHT Sponsored studies, the Research & Development department must be informed as the Sponsors representative.

The project is subject to the Research Governance Framework for Health and Social Care, 2nd Edition (DOH 2005) and if a CTIMP trial, The Clinical Trials Regulations and its subsequent amendments.

Please ensure that you are familiar with all ULHT Lincolnshire Clinical Research Facility SOP’s and comply with those relevant to your project. All current SOP’s are available at: http://www.ulh.nhs.uk/for_staff/lincollshire_crf/sop.asp

Please note that this Trust approval applies only to the documents listed above. Any changes to the protocol and/or study documents can only be initiated following notification to and approval by all relevant parties, such as the MHRA, Research Ethics Committee, R&D. All correspondence to the Ethics committee must be copied to Research & Development in order to maintain your Trusts Research & Development approval and indemnity status.

Please contact Dr. T Ahmed, Head of R&D & IP Lead/Director of LCRF, if you require any further information.

On behalf of the Trust, I wish you every success with the study.

Yours sincerely,

[Signature]

Dr Tanweer Ahmed
Head of Research and Development / Director of Lincolnshire Clinical Research Facility

CC: Academic Supervisor
Local Collaborator
Dear Ms Brough

Study title: Identifying developmental pathways of NEAD using MSFA
Chief Investigator: Jenna Brough
REC No: 14/WA/1214
Date of permission: 16 December 2014

List of all site(s) for which NHS permission for research is given:

Lincolnshire Partnership NHS Foundation Trust

NHS permission for the above research has been granted by Lincolnshire Partnership NHS Foundation Trust on the basis described in the application form, protocol and supporting documentation.

Permission is granted on the understanding that the study is conducted in accordance with the Research Governance Framework, ICH GCP and NHS Trust policies and procedures (available at http://www.lpt.nhs.uk/).

Permission is only granted for the activities for which a favourable opinion has been given by the REC [and which have been authorised by the MHRA]

List of any conditions of approval: N/A

The research sponsor or the Chief Investigator, or the local Principal Investigator at a research site, may take appropriate urgent safety measures in order to protect research participants against any immediate hazard to their health or safety.

The Research and Effectiveness office should be notified, at the address above, that such measures have been taken. The notification should also include the reasons why the measures were taken and the plan for further action. The Research and Effectiveness Office should be notified within the same time frame of notifying the REC and any other regulatory bodies.

Any research carried out by a Trust employee with the knowledge and permission of the employing organisation will be subject to NHS indemnity. NHS indemnity provides indemnity against clinical risk arising from negligence through the Clinical Negligence Scheme for Trusts (CNST). Further details can be found at Research in the NHS: Indemnity arrangements (Department of Health 2005).
All amendments (including changes to the local research team) need to be submitted in accordance with guidance in IRAS.

Please inform the Research and Effectiveness department of any changes to study status.

Please note that the NHS organisation is required to monitor research to ensure compliance with the Research Governance Framework and other legal and regulatory requirements. This is achieved by random audit of research.

We are pleased to inform you that you may now commence your research. Please retain this letter to verify that you have Trust permission to proceed. We wish you every success with your work.

Yours sincerely

Dianne Tetley
Assistant Director Research, Innovation and Clinical Effectiveness
Lincolnshire Partnership NHS Foundation Trust

Cc: Sponsor – University of Lincoln sowen@lincoln.ac.uk
Local Collaborator – Dr Tracey Swaffer tracey.swaffer@lpt.nhs.uk
Supervisors – ddawson@lincoln.ac.uk; mgresswell@lincoln.ac.uk; nmoghaddam@lincoln.ac.uk

Enc: Data Protection Guidance on the transportation of personal identifiable data
15th June 2015

Dr Sumcet Singhai
Nottingham University Hospitals
Neurology Department, Level D, West Wing
Queens Medical Centre
Derby Road
Nottingham
NG7 2UH

Dear Dr Sumcet Singhai

<table>
<thead>
<tr>
<th>Short Title / Acronym</th>
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</thead>
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<tr>
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<tr>
<td>Long Title</td>
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**PROJECT MILESTONES**

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<th>Recruitment Target</th>
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<tr>
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<td>Recruitment End Date</td>
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The R&I Department have considered the following documents submitted on 9/06/15 and there is no objection from the NHU R&I Office to the implementation of this amendment. The documents reviewed are detailed below:

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<th>Document</th>
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<td>06/06/2015</td>
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<tr>
<td>Participant Consent Form (Relative/Professional)</td>
<td>2.2</td>
<td>06/06/2015</td>
</tr>
</tbody>
</table>
The amendment may therefore be implemented immediately at this site under the conditions of the existing NHS Permission.

Please note that you may only implement changes that were described in the documents listed above.

Yours sincerely,

[Signature]

Dr. Brian Thomson / Dr. Maria Kourfalli
Director of R&D / Deputy Director Research and Innovation
Dear Dr. Sumeet Singhal,

<table>
<thead>
<tr>
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<tr>
<td>1st Patient to be Recruited by</td>
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</table>
The R&I Department has reviewed the following documents and NHS permission for the above research has been granted on the basis described in the application form, protocol, and supporting documentation. The documents reviewed were:

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</tr>
<tr>
<td>GP/consultant information sheet</td>
<td>1.0</td>
<td>19 Oct 2014</td>
</tr>
</tbody>
</table>

Your study now has NHS permission, on the understanding and provision that you will follow the conditions set out below.

**Conditions of Approval**

The Principal Investigator is responsible for:

1. Compliance with all relevant laws, regulations and codes of practice applicable to the trial including but not limited to, the UK Clinical Trials Regulations, Medicines for Human Use (Clinical Trial) Regulations 2004, principles of Good Clinical Practice, the World Medical Association Declaration of Helsinki entitled 'Ethical Principles for Medical Research Involving Human Subjects' (2013 version), the Human Rights Act 1998, the Data Protection Act 1998, the Medicines Act 1968, and the NHS Research Governance Framework for Health and Social Care (version 2 April 2005). Should any of these be revised and reissued this will apply. Copies of the up-to-date regulations are available from the R&I Office or via the R&I website [http://nuhri.se.org](http://nuhri.se.org).

2. Submission of study amendments to the Ethics committee and MHRA in accordance with the IRAS guidelines. Amendments and information with regards to changes in study status must be sent to R&I, (this includes changes to the local study team). Within 35 days from the receipt of a valid amendment submission, the R&I department will inform you if the amendment cannot be implemented locally. If no objections are raised NHS permission is valid and the amendment may be implemented.

When submitting documents for studies adopted into the NIHR portfolio please send the information to the Clinical Research Network: East Midlands (CRN:EM) ([CSP.CRNEMidlands@NIHR.ac.uk](mailto:CSP.CRNEMidlands@NIHR.ac.uk)). When submitting documents for all other studies please use the email address [rdamend@nuh.nhs.uk](mailto:rdamend@nuh.nhs.uk).
3. Ensuring all study personnel, not employed by the Nottingham University Hospitals NHS Trust hold either honorary contracts/letters of access with this Trust, before they have access to any patients or staff, their data, tissue or organs or any NUH facilities.

4. For initiating and delivering research in accordance with the Department of Health’s Plan for Growth. The first patient, first visit should occur within 70 days from the receipt of a valid submission in R&I. This applies to all studies where:

   i. The research is classed as a “clinical trial” on the IRAS filter page (first 4 categories)

5. Ensuring the research team via an identified individual, collaborates with the department of R&I and the CRN EM in reporting recruitment data using Documents and the CRN EM Study Tracker.

6. Ensuring that for GTAC-approved studies, the NHS permission is forwarded to GTAC via the sponsor. GTAC should then issue a site authorisation letter which must be received by each site prior to recruitment commencing. A copy of this letter must be forwarded to R&I.

7. Comply with requests from NUH R&I to allow monitoring of research to comply with the Research Governance Framework and other applicable regulations.

8. Record all types of adverse events (including Suspected Unexpected Serious Adverse Drug Reaction SUSARs) in the patient medical records and study documentation and report to the sponsor as required by the protocol.

9. Report any Serious Breach of the UK Clinical Trial regulations in connection with the trial or Serious Breach of the protocol, immediately after becoming aware of the breach to the study sponsor.

10. Reporting any changes to the study to R&I by letter or e-mail. These should not be implemented until agreed with R&I.

For NUH sponsored studies only, the Chief Investigator is responsible for:

i. All duties as detailed in the “Clinical Trial Delegation of Sponsorship responsibilities to Chief Investigator” agreement.

ii. Contacting the sponsor for review of all amendment documentation prior to submission to the HRA and MHRA. Please note that according to HRA and MHRA regulations, all submissions of amendments need to be signed by the authorised sponsor’s representative. All relevant documentation should be emailed to r&iamend@nhs.nhs.uk.

iii. Sending copies of the completed Annual Progress Reports, Development Safety Update Reports, and End of Study report required by the Ethics Committee and the MHRA (if appropriate) to the sponsor researchsponsor@nhs.nhs.uk.
iv. Notifying NUH R&I of all SAEs by completing and sending the “Serious Adverse Event reporting form” to R&I (only via fax, e-mail or by hand), within 24hrs of becoming aware of the event. Further guidance can be found in the R&I Adverse Event SOP (SOP-RES-019).

v. Reporting any Serious Breach of the UK Clinical Trial regulations in connection with the trial or Serious Breach of the protocol, immediately after becoming aware of the breach to NUH R&I as sponsor. Further guidance can be found in the R&I Non Compliance and Serious Breach Reporting SOP (SOP-RES017).

This approval letter constitutes a favourable Site Specific Assessment (SSA) for this site.

If you have any queries regarding the milestones or points detailed in this letter, please contact the Research Project Manager responsible for managing the performance of the study at NUH. This information is available on http://nuhrise.org.

Please note that the R&I department maintains a database containing study related information, and personal information about individual investigators e.g. name, address, contact details etc. This information will be managed according to the principles established in the Data Protection Act. Yours sincerely,

Dr Brian Thomson / Dr Maria Koudali
Director of Research and Innovation / Deputy Director Research and Innovation
Appendix B: Project protocol (version 1.2)

Using Multiple Sequential Functional Analysis (MSFA) to identify potential developmental pathways of Non-Epileptic Attack Disorder (NEAD)

Short title:
Identifying developmental pathways of NEAD using MSFA

Background and Rationale

Non-Epileptic Attack Disorder (NEAD)

Non-epileptic attacks can be succinctly described as abrupt episodes of altered behaviour which resemble epileptic attacks but are devoid of the characteristic clinical and electrographic features of epilepsy (Liske & Forster, 1964). Based on current understanding, when epilepsy and other medical causes are ruled out, such attacks are considered to be underpinned by psychological processes (Cuthill & Espie, 2005).

Non-Epileptic Attack Disorder (NEAD) is a diagnostic term for people who experience non-epileptic attacks (Betts & Boden, 1991), and is also known as Psychogenic Non-Epileptic Seizures (PNES). Through including the term ‘seizures’, PNES can be confusing for clients and clinicians, therefore a term not associated with epilepsy such as attacks or events may be preferable (LaFrance & Benbadis, 2006). Conversely, the term NEAD can technically encompass episodes/behaviour which have an organic aetiology but are non-epileptic, for example, syncope and dystonia (Gates, 2000 as cited in Benbadis, 2005). The minority of non-epileptic attacks are thought to have organic origins and these are often easily investigated, diagnosed, and treated (Locke, Berry, Fakhoury, & Schmitt, 2006 as cited in Binder & Salinsky, 2007), therefore non-epileptic attacks and NEAD will be the terms adopted from this point onwards to describe the previously defined behaviour and diagnosis.

NEAD has been estimated to affect between two and 33 people per 100,000 of the general population (Benbadis & Hauser, 2000), suggesting that up to 21,000 people in the UK may experience non-epileptic attacks. NEAD remains a diagnosis of exclusion, using video electroencephalography (EEG) data to rule out the presence of epileptic activity preceding, during, and after seizure-like episodes (Mostacci et al., 2011). However, to complicate accurate diagnosis further, research suggests that NEAD is co-morbid in up to 10% of people with diagnosed epilepsy (Benbadis, Agrawal, & Tatum, 2001; Martin et al., 2003). It is hypothesised that these people develop non-epileptic attacks after the onset of epilepsy through symptom modelling, a behavioural concept of learning through observation (Bautista, Gonzales-Salazar, & Ochoa, 2008).

Existing literature
NEAD has been recognised under various descriptions, for example, Gamgee in 1878 termed what would now be considered NEAD, hystero-epilepsy. More recently, non-epileptic attacks have been described as dissociative seizures (Brown & Trimble, 2000). The earliest explanation was psychodynamic theory, proposing that psychic conflict resulting from traumatic experiences is converted into physical symptoms to reduce anxiety and shield the conscious self from painful emotions (see Breuer & Freud, 1974). This appears based on the observation that clients who presented with non-epileptic attacks frequently reported traumatic histories (Devinsky, 1998).

In the 1980’s, later than the general shift in psychology, researchers began to move away from psychodynamic theory in favour of behavioural explanations (Ramani, Quesney, Olson, & Gumnit, 1980). Early behavioural theorists conceptualised non-epileptic attacks as learned behaviour, supported by observations that attacks were mainly found in people with experience (direct or observed) of epilepsy or similar altered states (Hopkins, 1989). Devinsky (1998) highlighted the influence of psychodynamic principles on behavioural theory, with relieving internal conflict proposed as a primary gain maintaining NEAD. The behavioural secondary gains described were, the support/care elicited by an attack and the avoidance of aversive situations.

In a review by Bodde et al. (2009), it was found that much research has focussed on identifying risk factors associated with NEAD, rather than on the refinement of a psychological theory to explain the presentation. Risk factors identified include: trauma (including abuse and neglect), evidence of borderline personality disorder, head injury, anxiety, inhibition of emotions, and stressful life events around the time of onset.

It can take an average of seven years of living with an epilepsy diagnosis, and related restrictions, before clients receive a revised NEAD diagnosis (Carton, Thompson, & Duncan, 2003; Reuber, Fernandez, Bauer, Helmstaeder, & Elger, 2002). This typically includes years of taking anticonvulsant medication which present the risk of toxicity and other side-effects (Liske & Forster, 1964; Reuber & Elger, 2003). Binder, Salinsky and Smith (1994) identified different psychological profiles in clients with NEAD and clients with epilepsy, which improved diagnostic accuracy from 74% using EEG data, to 81% using EEG data and the psychological profile (Storzbach, Binder, Salinsky, Campbell, & Mueller, 2000). However, Binzer, Stone and Sharpe (2004) suggest that while such profiles may support diagnosis, they are not distinct to clients with NEAD; rather they reflect differences commonly present in people with other psychologically underpinned phenomena. For example, a recent study found a statistically similar personality profile in people with NEAD and people with insomnia (Bodde et al., 2011). This suggests such profiles are only useful for supporting the NEAD/epilepsy differential diagnosis process and do not offer anything to improve understanding of the aetiology of NEAD.

Limitations of existing literature
Critically, much literature relating to NEAD is founded upon quantitative, cross-sectional research (see Bodde et al., 2009). A problem with reliance on cross-sectional designs, whereby NEAD and risk factors are measured simultaneously, is that it is difficult to determine whether these factors preceded or followed the onset of attacks. Such research has resulted in the identification of psychosocial factors more common in those with NEAD than in those with epilepsy, including; trauma (Rosenberg, Rosenberg, Williamson, & Wolford, 2000), childhood sexual and physical abuse (Alper, Devinsky, Perrine, Vazquez, & Luciano, 1993), head injury (Westbrook, Devinsky, & Geocadin, 1998) and family conflict (Wood, McDaniel, Burchfiel, & Erba, 1998). However, the factors identified are common across many other client groups and clinical populations. Additionally, risk factors are relatively common in the general population (e.g. trauma: Norris & Slone, 2013), yet NEAD is relatively rare. The ubiquity of such factors calls into question their individual predictive validity and explanatory utility and raises the question of how they interact to produce NEAD.

Baslet, Roiko and Prensky (2010) describe clients with NEAD as a heterogeneous group. This is supported by recommendations that treatment should be idiographic (LaFrance & Devinsky, 2002; Rusch, Morris, Allen, & Lathrop, 2001), with researchers proposing models of psychological formulation (Binzer et al., 2004; Reuber, 2009) to indicate appropriate intervention plans. Considering these recommendations it is unsurprising that previously employed nomothetic structural approaches, which seek to identify and describe features of phenomena, have failed to adequately conceptualise the complexity of NEAD. This indicates the urgency to explore whether a functional approach will offer more to understanding NEAD.

**The proposed research**

The proposed research aims to address the limitations of nomothetic, structural approaches by applying an idiographic functional approach to understanding how and why NEAD develops. Yin (1994) suggests ‘how’ and ‘why’ questions should be addressed through case study research.

Bromley (1990) describes a case study as “a systematic inquiry into an event or a set of related events which aims to describe and explain the phenomenon of interest” (p. 302). Despite criticism of case study research, that at best it provides interesting presentations of unique cases, Bromley (1986) proposes that being sensitive to uniqueness is a strength of case studies over cohort studies. By analysing cases individually researchers are able to modify initial conceptual frameworks in response to convergent and divergent features arising in new cases (Bromley, 1990).

Given the limitations of identifying risk factors using cross-sectional structural cohort studies, the proposed study aims to add to the understanding of NEAD by undertaking case study research to explore the development of non-epileptic attacks in client’s histories.

**Multiple Sequential Functional Analysis (MSFA)**
Functional analysis is a method which attempts to understand the function of behaviour by identifying variables which strengthen or reduce the likelihood of a specific behaviour occurring. A particular behaviour (or ‘target behaviour’) is understood through the use of an A:B:C: analysis. The A: stands for antecedent which is an event that occurs immediately prior to the B: which is the behaviour. The C: stands for the consequence which is the outcome of the behaviour (Sturmey, 2008). Functional analysis has a long history and is being increasingly used to advance understanding of a wide range of complex phenomena (Hanley, Iwata, & McCord, 2003), including: depression (Kanter, Cautilli, Busch, & Baruch, 2005), domestic violence (Bonem, Stanely-Klime, & Corbin, 2008), eating disorders (Slade, 1982), and self-injury (Bachman, 1972; Iwata, Dorsey, Slifer, Bauman, & Richman, 1994; Nock & Prinstein, 2004).

A particular type of functional analysis that has been used as a case study research methodology is Multiple Sequential Functional Analysis (MSFA: Gresswell & Hollin, 1992). MSFA aims to provide a framework for understanding the functional development of behaviour across the life of an individual. Using case material from multiples sources, a chain of A:B:C: functional analyses are developed, linked by the identification of key learning experiences which are hypothesised to have influenced the development of the target behaviour across time. This sequential analysis generates explicit hypotheses about the functional relationships between events and behaviour (Dawson & Gresswell, 2010; Gresswell & Hollin, 1992), and has been successfully used to facilitate understanding of the development of complex behaviour, including: multiple murder (Gresswell & Hollin, 1992), violent behaviour (Hart, Gresswell, & Braham, 2011), offence paralleling behaviour (Dawson & Gresswell, 2010) and female perpetrated intimate partner violence (Mappin et al., 2013). See Methodology and Data Analysis sections for a description of the MSFA process.

Utilising an idiographic case study approach, the research will aim to generate an in-depth understanding of the development of non-epileptic attacks that nomothetic approaches unable to achieve. The research will use MSFA which applies an established theoretical model, the behavioural model of operant learning (see Skinner, 1974), to attempt to understand the functional relationships between factors and behaviour.

**Clinical implications and relevance to clinical psychology**

A review by Bodde et al. (2009) suggests that, at present, there is no universally accepted psychological model to explain why NEAD develops and, therefore, there is no real understanding of what should be targeted in treatment or prevention. This research aims to contribute to the revival of theory development in this area, which has been identified as imperative for reducing the reliance on diagnosis through exclusion of epilepsy, and for informing targets for psychological intervention (Reuber, 2008).

Carton et al. (2003) found that receiving a NEAD diagnosis can be more distressing when clinicians lack a clear understanding of what NEAD is, and
therefore struggle to explain it adequately to clients, something they found to be common. They found an association between confusion, anger, and disagreement with a revised NEAD diagnosis and poorer prognosis (in terms of reduction in attack frequency and severity, and quality of life). This supports the need for further research into how non-epileptic attacks develop to improve clinical understanding and, potentially, client prognosis. This research may also demonstrate an acceptable method of developing and delivering a diagnosis using a robust theoretical framework.

Aims and Research Questions

The proposed research aims to use MSFA as a case study framework for examining the development of non-epileptic attacks in the individual life trajectories of a small group of adults with NEAD. The research aims to identify, examine, and compare and contrast these trajectories to generate hypotheses about the potential functions of non-epileptic attacks for these individuals, and synthesise new information which may contribute to theory development in this area.

The questions guiding the research are:

- How do non-epileptic attacks appear to develop in the histories of a sample of adults diagnosed with NEAD?
- What are the functions of non-epileptic attacks for these individuals?
- How do previously suggested risk factors appear to interact to influence the development of NEAD in these individuals?
- Are there similar pathways in the development of NEAD for the different individuals?
- Do the non-epileptic attacks have similar functional qualities for the different individuals?

Method

Methodology

The proposed study will use MSFA, a case study approach embedded in the methods, evidence base and philosophical assumptions of radical behaviourism (Gresswell & Hollin, 1992). MSFA was developed to provide a framework for understanding more complex behaviour where many environmental antecedents are identified as potential triggers, as appears to be the case with NEAD.

Developed by Gresswell and Hollin (1992), MSFA organises information into a series of A:B:C:s to account for complex chains of behaviour. It represents a developmental process whereby one A:B:C: explicitly influences the (A:) antecedents of the next, aiming to demonstrate the influence of
previous events on subsequent behaviour. In line with radical behaviourist principles, (B:) which stands for behaviour includes that which is overt (directly observable) and covert (thoughts, feelings and physiology). As with functional analysis, MSFA does not purport to make statements of causality, however, the ordering of complex material from multiple sources can lead to explicit hypotheses about causality based on the temporal relationships between variables. The (C:) which stands for consequences, are those which appear to function to strengthen or reduce relevant behaviour. A summary of learning as a result of each A:B:C: is hypothesised, to explain how the participant may have changed in their repertoire of behaviour as a result of the learning experience.

Criteria for conducting case study research

Bromley (1986) described criteria which must be met for case study research to be considered a worthwhile scientific enterprise:

5. It must give an **explanatory account** of the reasons for behaviour. The proposed research aims to produce an explanatory account of the development and maintenance of NEAD, underpinned by the behavioural principles of operant learning (Skinner, 1974).

6. It must aim to **improve knowledge** by providing new information which can be drawn on by future researchers. The proposed research aims to add to existing knowledge by using a method novel to explore the development of NEAD which may identify important new information to be examined in future research.

7. It must **develop or sustain the discipline** of studying individual cases. Applying MSFA to understanding NEAD will develop the discipline of studying individual cases by adding to the assessment of the utility of this research method.

8. Depend on **acceptable procedures** and arrangements. The procedure for MSFA is well-established and the more general research procedure will be considered through university and NHS boards of ethics, and supervision between the Chief Investigator (CI) and the research team.

Epistemological position

The epistemological position underpinning this research is functional contextualism (Gifford & Hayes, 1999). From this philosophical position, behaviour is understood within the context it occurs, and behaviour which is effective in meeting its intended consequences is considered pragmatically true (Fox, 2006). Within functional contextualism the aim of analysis is to identify rules and theories that are pragmatic to other researchers (Hayes, 1993). This research aims to predict the influence of events and psychosocial factors on the
development of NEAD. It will consider the function of participant behaviour (including verbal behaviour within interviews) within its context, and will make links between the behaviour and all available data, to achieve a coherent working understanding of the development and maintenance of non-epileptic attack behaviour for each participant.

Participants

Sample size

MSFA is an intensive methodology collecting a comprehensive amount of data from multiple sources. In line with other MSFA studies (e.g. Mappin et al., 2013), it is proposed that a minimum of three and a maximum of six participants will be recruited. Due to the intensive nature of the method, it is believed that this will be sufficient to capture a potential range of learning sequences/pathways to the development of NEAD.

Recruitment

Dr Sumeet Singhal (Consultant Neurologist) has agreed to support the recruitment to the study through his once weekly outpatient clinic in [name of clinic]. Dr Singhal will understand the inclusion and exclusion criteria for participants (later described). Eligible participants will be given the participant information sheet (Appendix A), and those who are interested will be asked to email or telephone the CI. The CI will answer any questions and provide more information about the research. If the participant would still like to take part the initial interview will be arranged at a convenient time and location.

Consent

At the beginning of the initial interview any further questions will be answered and informed consent to participate will be gained with a consent form being signed (Appendix B). Participants will need to consent to all elements of the study: one-to-one interviews, the CI accessing relevant files, and an interview with a relative/professional. A separate information sheet (Appendix C) and consent form (Appendix D) will be given to the identified relative/professional.

If the information sheet attracts eligible participants exceeding the maximum required, participants will be recruited in the order in which they have expressed an interest. Any remaining prospective participants will be contacted to let them know that the study no longer requires participants and they will be thanked for their interest.

Inclusion criteria

Identified prospective participants will be eligible for participation if they are 18 or over with a diagnosis of NEAD and are accessing services in the identified Trusts. Relatives/professionals must also be 18 or over.
Exclusion criteria

Participants and relatives/professionals will be excluded if they are unable to communicate and understand English spoken language. This is due to the in-depth nature of the interviews which comprises much of the study data. The constraints of the study budget would not allow for the expense of a translator/interpreter.

Participants who do not consent to their files being accessed will be excluded from the study due to the triangulation being a core element of the analysis. For the same reason, participants who cannot identify, or do not consent to, a relative/professional being interviewed, will also be excluded from the study.

Participation

It is proposed that participants will be seen by the CI for one-to-one interviews for between 5-7 hours in total, over multiple sessions. Interviews conducted with a relative/professional will last approximately 1-2 hours.

Dropout

A consecutive strategy will be employed, whereby recruitment will continue until either the maximum number of participants is reached, or the time scale of the study suggests there would be too little time to begin the process with another participant (and a minimum of three participants’ data has been obtained and they can no longer withdraw it from the study). This strategy should reduce the impact of any drop-out.

Procedure

<table>
<thead>
<tr>
<th>Interviews 1 and 2</th>
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<tr>
<td>Take full developmental history</td>
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| Organise data into chronological order and develop initial MSFA |

<table>
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<tr>
<th>Interviews 3 and 4</th>
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<tr>
<td>Gather more information relevant to developing MSFA</td>
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| Refine and update MSFA as appropriate |

<table>
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<tr>
<th>Triangulation</th>
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<tr>
<td>Relative/professional interview</td>
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</table>
Figure 1: The proposed procedure for each case.

**Interviews**

It is proposed that each participant will engage in up to four interviews, each of up to 90 minutes duration. The CI will only be aware of basic information about the participant and their experience of NEAD prior to the interview, this will include: name, age, how long since their NEAD diagnosis, and any previous diagnoses such as epilepsy.

The CI acknowledges that their previous experience working with clients with NEAD on placement will bring benefits, as well as issues, to the research. The CI will use their previous clinical experience of this client group to build rapport and demonstrate empathy, whilst holding in mind that they may have pre-conceived ideas about the experiences of people with NEAD.

At the first interview information about the study and procedure will be discussed again, and the participant will be given the opportunity to ask questions. Confidentiality limits will be outlined; that any disclosure of current risk to the participant’s or other’s safety may have to be reported. The potential for the interviews to evoke strong emotion and distress will be discussed, and a plan for the participant to access support through their current care network if required, will be agreed. At the end of each interview participant’s well-being will be discussed, and any identified issues will be considered and taken to the relevant professional if necessary.

The interviews will be engaging yet directive and the CI, as a Trainee Clinical Psychologist, will utilise therapeutic skills to build rapport. Interviews will be recorded on a Dictaphone and the CI will take notes relevant to developing the MSFA, which will not include any participant identifiable information. The interviews will not need to be transcribed as no textual analysis will be undertaken. The interviews will follow a semi-structured schedule focusing on taking a detailed clinical history for each participant. The schedule will be informed by factors associated with NEAD considered within a review by Bodde et al. (2009). Details from across all areas of the participant’s life will be sought
(for proposed interview themes, see Appendix E). The interview format will follow the principles of functional analysis assessment methods (see Sturmey, 2008), in order to gather data to generate a comprehensive conceptualisation of each case. The length of each interview will be determined by the CI identifying that a point has been reached where no new themes are emerging, and sufficient information has been gathered relevant to that stage of the process (see Figure 1.).

The CI will arrange a final one hour session to check the information gathered from all sources with the participant. This will involve checking out the order of events and asking for feedback in case anything is missing or doesn’t make sense (see Appendix E for more information).

Relative/professional interviews

Each participant will help identify an appropriate relative or professional, ideally someone who has known them for the longest and/or has had the most involvement with their experience of NEAD. Informed consent will be gained, and the aims of the study and confidentiality limits will be discussed before the interview begins. This interview will explore the relative/professional’s perspective on the development of NEAD in the participant’s history (see Appendix E). The interview will be audio recorded and anonymous notes made as necessary. It is acknowledged that the relative/professional may share information/opinions that the participant is not yet aware of. It is explained in the information and consent forms (Appendices C and D) that participants will review information gathered in a final session, therefore, they will give informed consent to information from their interview being shared with the participant.

File reviews

It is expected that there will be varying notes available depending on the participants’ involvement with services. Accessing files will also depend on how old they are and how they were recorded (paper/electronic). Additional participant consent may be required when requesting certain records. The procedure for accessing notes will be developed with the local collaborators and relevant NHS trusts.

Reflection

A reflective diary will be kept by the CI and completed after each interview/data collection session. It will be used to facilitative reflexivity and transparency in the research process by recording thoughts, assumptions, and subjectivities which may have influenced the process. This diary will only be seen by the CI and research supervisors, and no participant identifiable information will be recorded within it. The reflective diary will be analysed using MSFA in order to consider how learning experiences through the process may have influenced the CI’s subsequent behaviour.

Data Analysis

Each chronologically ordered individual narrative will be analysed using MSFA, linking a series of A:B:C sequences to identify which Consequences serve to
strengthen or reduce the behaviour of interest through reinforcement or punishment. Key learning as a result of the Consequences in one sequence creates the link to the Antecedent in the next A:B:C sequence (Gresswell & Hollin, 1992). With the arrow representing the learning, this is illustrated in Figure 2.

A:    B:    C:

A:    B:    C:

A:    B:    C:

Figure 2: an illustration of the sequencing in MSFA

The process will be considered complete when the CI and research supervisors determine that the life history and resulting MSFA has produced a theoretically coherent and complete understanding of the development of the participant’s non-epileptic attacks. The CI is a Trainee Clinical Psychologist with clinical experience of working directly with individuals with NEAD. They have significant clinical experience of assessing and formulating psychological difficulties (including NEAD) using various psychological models. The CI will work with the research supervisors who are qualified clinical psychologists to develop a comprehensive psychological explanation based within behavioural psychology. The participants will have the opportunity to give feedback on the gathered information from all of the sources with the chief investigator and will be encouraged to suggest any changes they feel are necessary in terms of order/missing information. The final MSFA may be edited based on the feedback provided by the participant in this session. It has been decided that participants’ will not be given the full explanation of the development of their non-epileptic attacks. This is due to the possibility that the participants may not be accepting of a psychological explanation at this time. Also, if they are not accessing psychological services currently and find it difficult to accept the explanation they may feel negative about accessing services as a result of this. It is also likely that if the participants do access psychology services in the future they will be working with a psychologist who utilises different theoretical models in their clinical work to that used in this research. This decision also helps to maintain the difference between this as a research project and any clinical experiences they may have had or go on to have in the future.

Some qualitative research methods have been criticised for merely producing a list of themes; Ayres, Kavanaugh, and Knafl (2003) advise that stand-alone themes have no explanatory power, without demonstrating how they work together data analysis is incomplete. The process of MSFA constitutes a within-case analysis as it will hypothesise the relationship between factors which has led to the development of non-epileptic attacks in the history of a participant.
Whilst considering the heterogeneity of the client group; across-case analysis may provide new understanding of the roles of suggested common factors in clients with NEAD. Once all cases are complete they will be reviewed for similarities and differences in: historical factors, reinforcement schedules, and functions. Utilising within- and across-case analyses, this research aims to enable the presentation of findings which allows readers to recognise individual experiences in a potentially generalisable way.

In the proposed study, data and method triangulation will be utilised. Triangulation aims to bring all data together to form a single comprehensive data set; in this case a comprehensive narrative of each participant's life with a specific focus on the events surrounding the development and maintenance of non-epileptic attacks. The triangulation process also means that discrepancies in the individual narratives/sources can be identified and may be resolved through considering all the sources (see Flick, 2004). If no consensus is reached through this form of checking, a best-fit approach related to the functional analysis can be taken, by considering information either side of the discrepancy chronologically and specifically discussing it with the research supervisors.

**Ethical Considerations**

The proposal for the study will be submitted to the University of Lincoln ethics committee and an NHS research ethics committee for approval. In addition it will be submitted to the relevant NHS Trusts for research governance approval.

The British Psychological Society (BPS) Code of Ethics and Conduct (BPS, 2009) and Code of Human Research Ethics (BPS, 2010) will be adhered to; principles relevant to the proposed study are as follows:

**Informed consent**

All participants will be fully informed of the nature of the research and will be given a detailed information sheet (Appendix A). Participants will be consenting to: one-to-one interviews, an interview with an agreed relative or professional (who will also need to provide informed consent), and a comprehensive file review. Participants will be encouraged to ask questions if anything is unclear or not explained.

**Withdrawal from the Study**

It will be emphasised throughout the process that participation is entirely voluntary. Participants are able to withdraw from the study at any point, and will be made aware that their interview data can be withdrawn up to two weeks after their final data collection interview with the chief investigator. BPS guidance suggests that participants should be allowed to withdraw their data before it is analysed. Although analysis actually begins after the second interview, it is triangulated with other sources after Interview 4. (see Figure 1.). Therefore this
has been judged as an appropriate cut-off for data withdrawal. Relatives/professionals can withdraw at any time and can withdraw their data up to two weeks after their interview as it is likely to be analysed and triangulated with the other sources after this point.

**Confidentiality and data protection**

Before consent is obtained and interviews begin, participants (and relatives/professionals) will be made aware of the confidentiality limits. The CI will make it clear that any identified/reported concern about the participant’s safety or the safety of others will be taken forward following the relevant trust and university policies.

Interview recordings will be transferred onto a secure laptop and erased from the Dictaphone at the first available opportunity. All paper records including notes from interviews and file reviews will be anonymous. Participants will be assigned a pseudonym at the beginning of their participation for differentiating and storing data, and for referring to interview excerpts in the final thesis and journal submission. Any need to transport data between secure laptops will utilise an encrypted memory stick.

Consent forms containing personal data will be stored securely in a locked cabinet in a locked office at the University of Lincoln. All data at all stages will be treated with strict confidence and will be accessible to the CI, supervisors and limited members of course staff only. All data related to the research will be stored for seven years after the completion of the study in accordance with university regulations and following this will be destroyed securely. Identifiable data will be destroyed securely three months following completion of the study.

**Protection of research participants**

The subject matter of the interviews carries the potential to cause distress in participants. The participant may be exposed to new potentially distressing information resulting from the data gathered from other sources discussed in the final session. The potential for distress will be discussed before consent is obtained, and before and after each interview session. It is hoped that the CI will be able to contain and manage moderate levels of distress. If any participant asks for or requires further support they will be referred to their current care network (Clinical Psychologist/GP) as appropriate. Consent to discuss any reported or identified distress with a current professional will be obtained.

The research supervisors, Dr Mark Gresswell, Dr Nima Moghaddan and Dr David Dawson will be available to contact for any concerns related to the CI or any element of the research.

To thank participants for their time and commitment to the study they will receive an inconvenience allowance (see Resources).
Debriefing of research participants

Participants will be provided with contact details for the CI and research supervisors. Participants will be offered the opportunity to receive an executive summary of the study following its completion.

Resources

The £500 research budget will be allocated as follows:

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<th>Description</th>
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<td>Participant inconvenience allowance</td>
<td>Up to 6 x (7 hours x £5 p/hour) in gift vouchers</td>
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<td>Travel expenses</td>
<td>24p per mile</td>
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<td>Stationery costs</td>
<td>2 x Black ink cartridges £12 each, paper, envelopes and stamps</td>
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<tr>
<td>Dictaphone</td>
<td>For interview recordings</td>
<td>£50</td>
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<td>Encrypted memory stick</td>
<td>For transporting data securely</td>
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Publication and Dissemination

This research study will be submitted in partial fulfilment of the requirements for the Trent Doctorate in Clinical Psychology (DClinPsy) in January 2016. It is also intended for submission to a peer-reviewed journal to further disseminate the findings. An executive summary of the research and findings will be offered to all participants including relatives/professionals.

Timescale

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<td>Feedback on research proposal</td>
<td>Identify research sites</td>
<td>Literature review</td>
<td>Develop MSFA skills</td>
<td>Develop and submit for ethical approval</td>
<td>Recruitment, Interviews and file reviews</td>
<td>Analysis of data</td>
<td>Write up thesis</td>
<td>Submit thesis</td>
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Word count: 5305
Appendix C: Recruitment Materials

Participant information sheet
06/06/2015 Version 2.4

Using Multiple Sequential Functional Analysis (MSFA) to identify potential Developmental Pathways of Non-Epileptic Attack Disorder (NEAD)

We’d like to invite you to take part in a research study. This research is being undertaken as an educational project. Joining the study is entirely up to you, before you decide we would like you to understand why the research is being done and what it would involve for you. This information will help you decide whether or not you would like to take part. Please feel free to talk to others about the study if you wish.

If you are interested in taking part please contact the researcher whose details can be found on the final page of this document. They will be able to answer any questions you may have. Speaking with them does not mean you have to take part; it is only to support you to make your decision with as much information as possible.

The first part of this sheet tells you the purpose of the study and what will happen if you take part. Then we give you more detailed information about the conduct of the study.

Thank you for your time.

What is the study about?

We aim to develop an understanding of the development of non-epileptic attacks in the histories of a small group of adults diagnosed with NEAD. We think this research is important because there are no substantial clinically useful understandings or explanations of how NEAD develops. As you may have found, many professionals lack a good understanding of what NEAD is and how to offer an explanation or support to people who receive this diagnosis. We will interview up to six adults, and an identified relative or professional for each person, and review relevant professional notes and files. We will use this information to attempt to produce an explanation of the development of each person’s attacks. Multiple Sequential Functional Analysis (MSFA) is a method of organising and analysing lots of complex information to try and understand the relationships between events and a particular behaviour. In this case we will use it to organise and analyse the relationships between life events which may have influenced the development of non-epileptic attacks.

You are being invited to take part in this study as you have been identified as an adult with a diagnosis of NEAD who is able to communicate in English.

Why is it important?
The purpose of this study is to add to the understanding of how NEAD develops. The current understanding of the development of NEAD is based on events/factors that people with NEAD possess or have experienced, but people with epilepsy have not. This does not explain why people develop NEAD and many people who have had these experiences do not have NEAD. We will look at your life and experiences to try and identify the pathway of how and why you developed non-epileptic attacks. Improving understanding of the development of NEAD will help us to guide future research which is greatly needed in this area. It may also help improve professional’s understanding so they are better able to support people who are diagnosed with NEAD, which is commonly a difficult time for people.

What would taking part involve?

If you decide to take part in the study and give your written consent, your main involvement would be interviews on a one-to-one basis. The interviews will be in private at a date and time convenient for you. The interviews will be audio recorded and the researcher will make some notes.

Participating in the study is likely to take between 5 and 7 hours of your time, although this will be split over up to 5 meetings. These meetings are likely to take place over a period of sixteen weeks (4 months). Variations of this schedule can be agreed individually with the researcher. The interviews will be scheduled to be as convenient as possible for you and will take place at either; a local NHS site, the University of Lincoln, the University of Nottingham, or your home.

The first two interviews will involve the researcher asking you lots of questions to get lots of information about significant events in your life and your experience of non-epileptic attacks. The third and fourth interviews will focus on filling in any gaps in the life history and checking things out/getting more details. The final session with the researcher will be a discussion of all of the information gathered which appears to be related to the development of your non-epileptic attacks. You will be encouraged to give your feedback and comments to the researcher.

Another part of the research involves the researcher interviewing a relative or professional. You will be asked to give written consent to this and identify together with the researcher who this will be. You will also be asked to give written consent for the researcher to review your case files including psychology and medical notes/reports relevant to your non-epileptic attacks and important life events/experiences. Looking at information from different sources will help us try to develop an understanding of your experiences and the factors which have been important in the development of NEAD in your life.

Participating in the research will not affect your current involvement with services and the researcher will be as flexible as possible to meet at dates and times most convenient for you. Also, if you decide not to take part your current involvement with services will not be affected, your participation is entirely optional. If you are already involved in any research it is important to let the
researcher know so you can discuss if it would be ok to participate in this research too.

As a token to acknowledge the significant time commitment required to participate, you will be given a £5 high street gift voucher per hour of time you spend with the researcher; as this will take up to 7 hours this will be a maximum of £35.

What are the possible benefits of taking part?

It is hoped that by taking the time to share your experiences we will develop a better understanding of the events/circumstances in your life that have influenced the path to where you are now. Understanding how NEAD develops will be useful for informing future research. This is needed as there are currently no substantial explanations for how and why NEAD develops. Contributing towards the development of such explanations is a worthwhile task as it may enable the identification of potential treatments and support better explanations when clients are diagnosed.

What are the possible disadvantages and risks of taking part?

The interviews will involve talking about many events in your life, positive and negative, past and current. This will include talking about things which you felt were negative experiences or things which were or are distressing for you including; traumatic experiences e.g. abuse or violent acts, bereavements, illnesses, and accidents. You do not have to answer questions if you don’t want to and you don’t have to give a reason. If you feel too distressed at any time you can stop the interview and you can discuss with the researcher whether there is the need to let those involved in your care know about it. In order to support this if it is needed, your GP will receive a standard letter informing them that you are participating in the study. At the end of each interview you will discuss how you are feeling with the researcher and you can both decide if you need any extra support. In the final session the researcher will share information gathered from your interviews, the interview with your relative/professional, and information from relevant files. This may include new and potentially distressing information that you may or may not agree with. You will be encouraged to share your opinions on the information and you will be able to access support for any distress this may cause. Also, it is important to consider that this study requires a significant time commitment from you.

How will my information be kept confidential?

All information collected about you will be kept strictly confidential, unless something you say suggests that you or someone else is or has been at risk of harm (this follows standard NHS procedures). Should such an issue arise; the researcher will try to discuss this with you.

You will be given a false name to protect your identity at the beginning of the research which will be used to separate and store all of your interview recordings and notes made. This false name will also be used in the written
research report and any publication of the study which may include interview quotes. The audio recordings of interviews will be transferred onto a password protected computer as soon as possible after each interview. The notes will be transferred to a locked file as soon as possible after being made.

The consent form you sign and your contact details will be stored securely at the University of Lincoln, separately from the information you give in interviews and information noted from your records. All of the information using the false name (interview recordings and notes made) will be stored in a locked cabinet or on a password protected computer.

All paperwork and information related to your participation in the study may be accessed by the researcher, researcher supervisors (Dr Mark Gresswell, Dr Nima Moghaddam, and Dr Dave Dawson) and administrators at the University of Lincoln. Access will only be granted if it is relevant and necessary to support the completion of the research. If the regulatory bodies within the NHS and the university need to check that the research is following the right procedures and policies they may need to see all or some of this information too.

**What will happen if I don't want to carry on with the study?**

You have the right to withdraw from the study at any time. You have the right to end the interviews and you can withdraw your data up to two weeks after your last interview, usually the fourth (not your final feedback meeting with the researcher). You should inform the researcher as soon as possible if you change your mind about taking part. You do not have to give a reason and your access and involvement with any services will not be affected.

**What happens after the study?**

The study will be submitted as part of a thesis for a Doctorate in Clinical Psychology at the University of Lincoln. The study may also be written up and submitted for publication in a scientific journal or the findings presented at conferences. You will not be identified in any presentation of the study or data (as false names will be used). All data related to the study will be held securely for 7 years at the University of Lincoln. Data containing your personal details will be held securely for 3 months and will then be securely destroyed.

You can ask to receive a summary of the overall study when it is completed.

**What if there is a problem and I want to complain?**

If you wish to complain about any element of the study, in the first instance please discuss your concerns with the researcher. If you remain unhappy or you would rather speak to someone else, complaints can be directed to the research supervisor:

Dr. Mark Gresswell  
DClinPsy  
School of Psychology
Brayford Campus  
University of Lincoln  
Lincoln LN6 7TS

or to the chair of the School of Psychology Ethics Committee:

Patrick Bourke  
Senior Lecturer in Psychology  
School of Psychology  
Brayford Campus  
University of Lincoln  
Lincoln LN6 7TS  
pbourke@lincoln.ac.uk

If you remain unsatisfied complaints can be directed to your local Patient Advice and Liaison Service (PALS).

Who has reviewed this study?

This study has been reviewed and has met with the approval of the University of Lincoln and the Wales REC4 NHS reviewing body. Permission has also been granted by the NHS trusts you access services through to undertake the research.

What do I do now?

If you are interested in being involved in the study please contact Jenna Brough by email 13451652@students.lincoln.ac.uk or by telephone 07437618228. Jenna will be able to discuss the research in more detail and answer any questions you may have.

If you would like to and are able to take part we will then arrange a time and place for the initial interview where written consent will be required.

Contact details

Jenna Brough, Trainee Clinical Psychologist (13451652@students.lincoln.ac.uk)  
Trent Doctorate in Clinical Psychology, College of Social Science, University of Lincoln, Bridge House, Brayford Pool, Lincoln, LN6 7TS.

Under the supervision of Dr Mark Gresswell (m.gresswell@lincoln.ac.uk), Dr Nima Moghaddam (n.moghaddam@lincoln.ac.uk) and Dr Dave Dawson (d.dawson@lincoln.ac.uk).

Thank you for your time
CONSENT FORM

Title of Project: Using Multiple Sequential Functional Analysis (MSFA) to identify potential developmental pathways of Non-Epileptic Attack Disorder (NEAD).

Name of Researcher: Jenna Brough

Assigned participant pseudonym:

Please initial box

1. I confirm that I have read the participant information sheet dated 06/06/2015 (version 2.4) for the above study. I have had the opportunity to consider the information, ask questions and have had these answered satisfactorily.

2. I understand that I will be assigned a false name (pseudonym) for the purposes of the research which will be used to differentiate and store my data and will be used in the written report and any published papers to protect my identity.

3. I give permission for my interviews to be audio recorded and understand that quotes may be used in the written report of the research and any published papers.

4. I understand that my participation is voluntary and that I am free to withdraw at any time without giving any reason, without my care or legal rights being affected. Furthermore, I understand that if I want to remove my data from the study I must do this within two weeks of the final data collection interview (not the final information checking meeting).

5. I understand that information I give and data collected in the study may be looked at by the following people: the researcher, research supervisors, administrators at the University of Lincoln and staff from regulatory bodies, where it is relevant to my taking part in this research. I give permission for these individuals to have access to my records for the purposes of this research and for ensuring procedures and policies are being followed correctly.
6. I give permission for the researcher to access my medical and social care records for the purposes of this research and understand that I may need to sign further forms to support the researcher to access these records.

7. I give permission for the researcher to inform my GP (via letter) that I am participating in this research study.

8. I agree to take part in all components of the above study detailed in the participant information sheet dated 06/06/2015 (version 2.4).

________________________  ______________________  ______________________
Name of Participant        Date                      Signature

________________________  ______________________  ______________________
Name of Person taking consent Date                      Signature
Using Multiple Sequential Functional Analysis (MSFA) to identify potential developmental pathways of Non-Epileptic Attack Disorder (NEAD).

We’d like to invite you to take part in a research study. This research is being undertaken as an educational project. Joining the study is entirely up to you, before you decide we would like you to understand why the research is being done and what it would involve for you. This information will help you decide whether or not you would like to take part. Please feel free to talk to others about the study if you wish.

Thank you for your time.

What is the study about?

We aim to develop an understanding of the development of non-epileptic attacks in the histories of a small group of adults diagnosed with NEAD. We think this research is important because there are no substantial clinically useful understandings or explanations of how NEAD develops. Many professionals lack a good understanding of what NEAD is and how to offer an explanation or support to people who receive this diagnosis. We will interview up to six participants, an identified relative or professional for each participant, and review relevant professional notes and files. We will use this information to attempt to produce an explanation of the development of each participant’s attacks. Multiple Sequential Functional Analysis (MSFA) is a method of organising and analysing lots of complex information to try and understand the relationships between events and a particular behaviour. In this case we will use it to try to organise and analyse the relationships between life events which may have influenced the development of non-epileptic attacks.

You are being asked to take part as a participant in this study, has identified you as a person who knows/has known them well in a personal or professional capacity. You have been identified as being over 18 and able to communicate clearly in English.

Why is it important?

The purpose of this study is to add to the understanding of how NEAD develops. The current understanding of the development of NEAD is based on events/factors that people with NEAD possess or have experienced, but people with epilepsy have not. This does not explain why people develop NEAD and many people who have had these experiences do not have NEAD. We will look at the life and experiences of up to six adults who have NEAD to try and identify the pathway of how and why they developed non-epileptic attacks. Improving understanding of the development of NEAD will help us to guide future research which is greatly needed in this area. It may also help improve professional’s understanding so they are better able to support people who are diagnosed with NEAD, which is commonly a difficult time for people.
What would taking part involve?

If you decide to take part in the study and give your written consent, you would be consenting to being interviewed by the researcher to give your perspective on the development of non-epileptic attacks in the life history of the participant you know.

Interviews will last between one and two hours and will be audio recorded. The researcher will also take notes during the session. A date and time convenient for you will be arranged for the interview session and it will take place at either; a local NHS site, the University of Lincoln, the University of Nottingham, or your home.

You do not have to take part but the methodology employed in this study relies on the ability to collect and compare information from different sources; the participant, a person who knows them well (you), and relevant medical/psychology files. This will then be used to attempt to produce a comprehensive individual explanation of the development of the participant’s non-epileptic attacks. It is important to note that information from your interview may be shared with the participant or identified by them in the research summary or any publications.

What are the possible benefits of taking part?

It is hoped that by taking the time to share your perspective we will develop a better understanding of the events/circumstances that have influenced the development of NEAD in the person you know well. Understanding how NEAD develops in up to six adults in this study will be useful for informing future research. This is needed as there are currently no substantial explanations for how and why NEAD develops. Contributing towards the development of such explanations is a worthwhile task as it may enable the identification of potential treatments in the future and support better explanations when clients are diagnosed.

What are the possible disadvantages and risks of taking part?

The interview will involve the researcher asking questions to get lots of information about events in the life of the person you know and your perspective on their non-epileptic attacks. This may include talking about things which may have been distressing/upsetting for the person you know well and maybe for you. You do not have to answer questions you don’t want to and you don’t have to give a reason. If you feel too distressed at any time you can stop the interview. At the end of the interview you will discuss how you are feeling with the researcher and you can both decide if you need any extra support. Also, it is important to consider that this study requires a significant time commitment from you.

How will my information be kept confidential?
All information collected will be kept strictly confidential, unless something you say suggests that you or someone else is or has been at risk of harm (this follows standard NHS procedures). Should such an issue arise; the researcher will try to discuss this with you.

The person you know well will have been given a false name to protect their identity at the beginning of the research. You will be referred to as their relative or professional for example “Jane’s relative” and therefore will not be identified in the storage of data or the write-up of the research. The written report of the research and any publication of the study may include quotes from your interview but this protection to your identity will apply.

The consent form you sign and your contact details will be stored securely at the University of Lincoln, separately from the information you give in the interview and the notes made. All of the information identified using the false name (interview recording and notes made) will be stored in a locked cabinet or on a password protected computer.

All paperwork and information related to your participation in the study may be accessed by the researcher, researcher supervisors (Dr Mark Gresswell, Dr Nima Moghaddam, and Dr Dave Dawson) and administrators at the University of Lincoln. Access will only be granted if it is relevant and necessary to support the completion of the research. If the regulatory bodies within the NHS and the University need to check that the research is following the right procedures and policies they may need to see this information too.

What will happen if I don't want to carry on with the study?

You have the right to withdraw from the study at any time and you do not have to give a reason. You have the right to end the interview and you can withdraw your data up to two weeks after it has been collected. You should inform the researcher as soon as possible if you change your mind about taking part.

What happens after the study?

The study will be submitted as part of a thesis for a Doctorate in Clinical Psychology at the University of Lincoln. The study may also be written up and submitted for publication in a scientific journal or the findings presented at conferences. You will not be identified in any presentation of the study or data (as false names will be used). All data related to the study will be held securely for 7 years at the University of Lincoln. Data containing your personal details will be held securely for 3 months and will then be securely destroyed.

You can ask to receive a summary of the overall study when it is completed.

What if there is a problem and I want to complain?

If you wish to complain about any element of the study, in the first instance please discuss your concerns with the researcher. If you remain unhappy or you
would rather speak to someone else, complaints can be directed to the research supervisor:

Dr. Mark Gresswell  
D ClinPsy  
University of Lincoln  
Brayford Pool  
Lincoln  
LN6 7TS

or to the chair of the School of Psychology Ethics Committee:

Patrick Bourke  
Senior Lecturer in Psychology  
School of Psychology  
Brayford Campus  
University of Lincoln  
Lincoln LN6 7TS  
pbourke@lincoln.ac.uk

If you remain unsatisfied complaints can be directed to your local Patient Advice and Liaison Service (PALS).

Who has reviewed this study?

This study has been reviewed and has met with the approval of the University of Lincoln and the Wales REC4 NHS reviewing body. Permission has also been granted by the relevant NHS trusts to undertake the research.

What do I do now?

If you are interested in being involved in the study please tell the person who identified you as their relative/professional. Give them permission to pass on your contact details and we will get in touch with you (usually within one week). Over the phone we will be able to discuss the research in more detail and answer any questions you may have.

If you would like to and are able to take part we will arrange a time and place for the interview where written consent will be required.

Contact details

Jenna Brough, Trainee Clinical Psychologist (13451652@students.lincoln.ac.uk)  
Trent Doctorate in Clinical Psychology, College of Social Science, University of Lincoln, Bridge House, Brayford Pool, Lincoln, LN6 7TS.  
Under the supervision of Dr Mark Gresswell (m.gresswell@lincoln.ac.uk), Dr Nima Moghaddam (n.moghaddam@lincoln.ac.uk) and Dr Dave Dawson (d.dawson@lincoln.ac.uk).

Thank you for your time
RELATIVE/PROFESSIONAL CONSENT FORM

Title of Project: Using Multiple Sequential Functional Analysis (MSFA) to identify potential developmental pathways of Non-Epileptic Attack Disorder (NEAD).

Name of Researcher: Jenna Brough

Assigned pseudonym: 

1. I confirm that I have read the relative/professional information sheet dated 06/06/2015 (version 2.2) for the above study. I have had the opportunity to consider the information, ask questions and have had these answered satisfactorily.

2. I understand that I will be assigned a false name (pseudonym) for the purposes of the research which will be used to differentiate and store my data and will be used in the written report and any published papers to protect my identity.

3. I give permission for my interview to be audio recorded and understand that quotes may be used in the written report of the research and any published papers.

4. I understand that my participation is voluntary and that I am free to withdraw at any time without giving any reason. Furthermore, I understand that if I want to remove my data from the study I must do this within two weeks of it being collected.

5. I understand that information I give may be shared with or seen by the participant.
6. I understand that information I give and data collected in the study may be looked at by the following people: the researcher, research supervisors, administrators at the University of Lincoln and staff from regulatory bodies, where it is relevant to my taking part in this research. I give permission for these individuals to have access to my records for the purposes of this research and for ensuring procedures and policies are being followed correctly.

7. I agree to take part in all components of the above study detailed in the participant information sheet dated 06/06/2015 (version 2.2).

________________________  __________________________  __________________________
Name of Participant        Date                        Signature

________________________  __________________________  __________________________
Name of Person taking consent Date                        Signature
Dear Dr <Name>,

Re: <Patient Name, Date of Birth, and Address>.

**Using Multiple Sequential Functional Analysis (MSFA) to identify potential developmental pathways of Non-Epileptic Attack Disorder (NEAD)**

I am writing to inform you that your patient has agreed to participate in the above research study.

The purpose of the study is to develop an understanding of the pathways of non-epileptic attack development in the histories of a small group of adults diagnosed with NEAD. We think this research is important because there are no substantial clinically useful explanations of how NEAD develops. Understanding more about the development will help direct further research and may improve professional’s understanding.

Your patient has been asked to participate because they are an adult with a diagnosis of NEAD, with the ability to communicate and understand spoken English. They were identified through attending Dr Singhal’s outpatient neurology clinic at [redacted]. Your patient will engage in in-depth interviews with myself which will involve gathering information about significant events in their life and their experience of non-epileptic attacks. They will also consent to a relative or professional being interviewed and the researcher reviewing medical and social care files relevant to their non-epileptic attacks and potentially related events.

I have enclosed a copy of the Participant Information Sheet for your reference, however if you have any queries or require further information please do not hesitate to contact me through the secure NHS.net email system: jenna.brough@nhs.net.

Yours sincerely,

Jenna Brough
Trainee Clinical Psychologist
(Chief Investigator)
Encs: Participant Information Sheet, version 2.4 dated 06/06/2015
Appendix D: Interview Guide

Interview Guide

Version 1.1
26/01/2015

Identifying Developmental Pathways of NEAD using MSFA

*Each interview will begin and end with the following prepared statements*

**Opening statement:**
Thank you for agreeing to be interviewed. Do you have any questions before we begin today? How are you feeling about the interview? Let me know if you need to take a break or if you are finding anything too distressing to talk about. I would just like to check again – are you happy for us to audio-record this interview? Do you feel ok to begin?

**Closing statement:**
Thank you again for your time. Is there anything you feel we haven’t covered in the interview or anything you would like to clarify? Is there anything else you would like to ask about the study? How are you feeling after speaking with me today? (prompt to discuss possible distress to identify need to signpost for further support from current professionals or GP).

*The sections below outline topics areas and provide some example questions.*

**Interview 1:**

Their understanding and experience of their non-epileptic attacks and their current situation

Tell me about your experience of non-epileptic attacks…
What do you understand these experiences to be about?
How has NEAD impacted your life? And the life of others?
Tell me about the last time you had an attack… (What was happening before? How were you feeling? Where were you? Who was there? What happened/what did people say happened? What happened afterwards?)

Tell me about the first attack you remember… (as above) What was happening in your life more generally around this time?

Typical attacks – frequency, how long they last, usual contexts, possible triggers, typical emotions, presence/absence of others, actual behaviour during, consequences/impact, mediating factors – times when it has been better or worse.

What has your involvement been with services (medical and psychological) since your attacks began? (further prompts regarding diagnoses, treatment, acceptance/disagreement with professional opinion/diagnoses)

Tell me about your life at the moment…
This will include prompts regarding employment, living situation, important relationships (partners, children, parents etc.), physical and mental health including current/ongoing stress.

Interview 2:

Full developmental history

Last time we talked about your attacks and how your life is at the moment. Today it would be useful if we could talk about your early life more.

What do you know about your birth and the family situation when you were born? (include prompts about living circumstances, medical concerns at birth) Tell me about your parents… who cared for you the most? Did your parents work?
Tell me about your relationship with any siblings… (including birth order, closeness, competition, development)
Tell me about your grandparents…
How was it growing up in your family?
Tell me about your family’s health and well-being….
Tell me about your experience of school (including social and academic experiences)
Tell me about your friends growing up
What would you say was the most significant relationship in your childhood?
(ask about the opposite)
Are there any times during your childhood that particularly stand out to you as being important or memorable?
Would you say there is anything in your childhood that you would regard as traumatic or distressing? (If yes) Can you tell me about that?
And going back to the first attack you remember (if in childhood), when was this in relation to what we have discussed today? What changed for you after that point?

**Interview 3:**

**Risk factors of interest and further information**

*Anything that didn’t fit in the first two interviews will be explored in interview three.*

*This interview will also explore risk factors of interest identified in previous interviews in more detail e.g. trauma, conflict, psychiatric history, stress at time of onset.*

*Also there will be a focus on how the onset and maintenance of attacks fits into the developmental history, potentially making links between important events/risk factors and the attacks chronologically.*

*This session will also focus on more intra-personal characteristics relating to personality, coping, and emotions.*

Tell me about how you cope with general stress…
Tell me about how you have responded/dealt with negative events e.g. bereavements, break-ups, arguments…
We all have worries, tell me what you worry about? Have these sorts of worries been long-standing for you?
How do you feel about your attacks?
How do you cope when things don’t go to plan?
When faced with problems how do you usually approach them?
What would you say are your strengths and weaknesses?

Interview 4:

**Clarifying, checking out and filling the gaps**

*This interview will check out and clarify anything that wasn’t clear from the previous interviews in order to ensure the chronology and relationship between events etc. is correct.*

*Where gaps have been identified in a participant’s history further questions will be asked to ensure there is a full history. E.g. You talked a lot about primary school but less about secondary school, tell me more about that if you can….*

Relative/professional Interview:

*The relative/professional interview will take place after the four participant interviews. They will be asked for their perspective on the experiences they have witnessed/been part of in the participant’s life.*

*The sections below outline topics areas and provide some example questions.*

Tell me about your understanding of X’s non-epileptic attacks ….

Have you ever witnessed them having an attack? (If yes) - Thinking about the most recent attack you witnessed, what was happening before? How did they
appear to be feeling? Where was it? Who else was there? What happened during the attack (behaviour, duration)? What happened afterwards?

(If they have witnessed more attacks) - Tell me about the first attack you remember witnessing… (as above)
Do you remember anything important/memorable happening in their life or the family’s life more generally around this time?
Do you recall any periods of time when their attacks have been better or worse?
Was there anything particularly memorable/important happening during this period?

What is your understanding of their non-epileptic attacks? What have you based this on? Have you communicated this to X? (If yes) How did they respond to this?

Tell me how you feel X copes with stress…
Tell me how X seems to deal with negative events e.g. bereavements, break-ups, arguments…
What would you say are X’s strengths and weaknesses?

(If a professional) When did you become involved with X? What has your role been (assessment, diagnosis, intervention)? Tell me about how X has engaged with your service…

(If a relative) Tell me about your relationship with X…
Tell me about any important events in the family history which X has been involved in or aware of….

*Depending upon who the relative is further questions may be asked about the family home, school experiences and childhood of the participant (similar to examples including in Interview 2 above.*
With the permission of the participant the relative/professional will be asked about their understanding of/perspective on identified risk factors e.g. trauma, psychiatric history, stress at the time of onset.

If gaps remain in the participant’s history the relative/professional will be asked if they have any knowledge of this time in the participant's life.

**Final checking session with participant:**

The researcher will share the relevant information in chronological/developmental order with the participant and the participant will be given the opportunity to give their opinion on the incorporated information from all sources. If the participant is not forthcoming with feedback the following prompts/questions may be asked.

What are your thoughts about the order of things?
Do you recognise the information as your experiences?
Is there anything you don’t recognise?
How do you feel about the information all together?
Is there anything that doesn’t make sense?
Is there anything important you feel has been missed?
Is there anything that you don’t agree with?
Tell me what you would change about it…
Identifying developmental pathways of NEAD using MSFA

Jenna Brough1,2, David M Gresswell1, Nima G Moghaddam1, David L Dawson1
1Trent Doctorate in Clinical Psychology
#Email: jennabrough@live.co.uk

**Background**

Non-epileptic attacks (NEAs) resemble and are often mistaken for epileptic seizures. However, Non-Epileptic Attack Disorder (NEAD) seems underpinned by psychological rather than neurological processes1. Due to the resemblance, much research has focused on identifying psychosocial factors differentiating the two populations, improving diagnostic accuracy2. Understanding of NEAD development is largely based on these factors, though they are common in other clinical3 and community populations. Examining how these factors interact in NEAD development may improve understanding. As structural research cannot explain such interactions/processes, a functional approach is indicated.

**Research Questions**

- How do NEAs appear to develop in the histories of a sample of adults with NEAD? (and are the developmental pathways similar?)
- What are the functions of NEAs for these individuals? (and are they similar?)
- How do previously suggested risk factors appear to interact to influence the development of NEAD in these individuals?

**Method**

The research used MSFA in a series of three case studies.

For each participant the following data was collected:

- 7 hours of participant interviews.
- 45-90 minute interview with a relative.
- Review of relevant files e.g. neurology, psychology and psychiatry.

**Multiple Sequential Functional Analysis (MSFA: Gresswell and Hollin4)**

MSFA organises information from multiple sources into a series of A:B:C:s to account for complex behaviour. It represents a developmental process whereby one A:B:C explicitly influences the (A:) antecedents of the next, demonstrating the influence of learning on subsequent behaviour. In line with radical behaviourist principles, (B:) includes overt (directly observable) and covert (thoughts/feelings/physiology) behaviour. The consequences (C:), are what appears to strengthen or reduce the behaviour in future.

**Results**

Jayden* did not develop adaptive strategies for dealing with social and emotional situations. Early illness reporting and post head injury seizures resulted in withdrawal and increased care. As seizures were treated NEAD appeared to develop. When expressing anger was punished, NEAs were generalised to enabling avoidance of anger evoking stimuli and internal anger.

Susan was punished for expressing negative emotion. Early dissociation and an incident of syncope were reinforced through avoiding (feared) punishment. NEAs appeared to develop in response to similar emotions in adolescence. A TIA triggered an increase in attacks due to increased emotionality and positive reinforcement.

Daisy functioned well under stress, until she had her children and continued to work excessively leading to a functional stroke. An earlier virus-related blackout when stressed and unwell was the only behaviour in her learning history that had reduced stress, NEAs seemed to develop in response to stress and increased symptoms.

**Discussion**

- Learning through observation or experience of epilepsy (symptom modelling5) applied to another altered state(syncope), with mirrored topography in each case.
- A limited behavioural repertoire, altered states with positive consequences, and later similar contexts, underpinned the development and maintenance of NEAD.
- Study limitations include inability to access historical files and that hypotheses were not verified (the functional analyses were descriptive6).
- Future research should explore if hypothesised processes are similar in others with NEAD and seek to verify hypotheses in intervention studies.
- These findings offer an understanding of potential mechanisms in NEAD development to inform the development of theory and specific treatment approaches.

*Pseudonyms have been used to maintain participant anonymity.

**References**