Psychological interventions for psychogenic non-epileptic seizures: A meta-analysis

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1. Introduction

Psychogenic non-epileptic seizures (PNES) have a debilitating impact on quality of life. This may involve psychological, social, financial and physical consequences including the inability to work, drive or carry out everyday tasks [1,2]. Despite the growing amount of research contributing to our understanding of PNES and its causes, there is little evidence available about successful treatments [3,4].

Prognosis for people with PNES is poor [5]. Diagnosis is often focused on the exclusion of epilepsy and consequently, PNES becomes a non-disease [6]. People with PNES tend to be marginalized between neurology and psychiatry, with neither profession taking ownership of patient care [7]. As such, many patients are not referred to or do not engage with mental health services [3,4,6]. Once a diagnosis of PNES is made, anti-convulsant therapy is typically ceased and treatment options are unclear and rarely pursued [1,7]. Stigma often surrounds a diagnosis of PNES, fueled by poor understanding, education or support for the condition [1,6]. Research also tells us that, without treatment, the majority of people with PNES continue to have seizures and many experience a worsening of symptoms [8,9,5].

Whilst PNES is a condition defined by physical manifestations, it is understood to be psychological in nature with a wide variety of aetiological factors involved [10,4,11]. Consequently, PNES represent a serious problem for clinicians in developing and implementing evidence-based psychological interventions and there is currently little in the way of quality evidence which can inform clinical treatment decisions [12]. The body of research indicates that psychological interventions for PNES are in the early stages of development. These encompass a number of approaches, the most
common being cognitive behavioral therapy (CBT), psychoanalyti-
cal and psychoeducational therapies.

The majority of the research into psychological interventions
for PNES is comprised of observational studies, involving pre-post
studies without control groups [12]. Most are small in scale
and conducted in hospital or medical facilities, reflective of real life
clinical treatment settings [13,14,9,15]. These studies are inter-
spersed with a handful of small scale and pilot randomized
controlled trials (RCTs) [16,17,18] providing a promising start in
the pursuit of high quality research into PNES interventions. On their
own however, they are insufficient in number to allow clinicians to
draw conclusions about broader treatment recommendations
[3,12,4].

There are several reasons for the limited number of high quality
studies in this field. RCT’s are typically performed in highly
controlled environments where extraneous variables can be
controlled. The majority of RCT’s do not allow for the presence
of co-morbid disorders, common among people with PNES [19],
leaving these people unrepresented in the research. RCT’s also
commonly require a single standardized treatment which is
difficult to develop for such a group as diverse as those with PNES
which can be a symptom of various affective and psychiatric factors [10,4]. Furthermore, RCT to waiting list or treatment as
usual (TAU) is unattractive, and can be deemed unethical for
patients who are unwell, when similar treatments are available
outside of the research setting [20].

Overall, the individual research studies are suggestive of
favorable outcomes in terms of reducing seizures for those who
complete psychological treatment. However, as a whole, the
literature is laborious to interpret as studies are often published in
a variety of different medical, psychological or psychiatric journals,
use different methodologies, and are presented in such a way as to
make them difficult to compare with one another. As it stands, the
evidence is indicative of both the current state of clinical
interventions for PNES as they occur in practice, and reflective of
the populations they aim to treat. It is also representative of the
diversity of approaches required in addressing such a heteroge-
neous group of patients and presentations [10,4,11,15]. The
observational designs utilized by the majority of researchers in
this field, whilst of limited methodological quality, have the
capacity to evaluate treatment outcomes in people with multiple
problems, complex or atypical presentations in real life clinical
settings [21]. Naturalistic studies inform clinicians, researchers
and other health professionals about treatments, as they would be
performed in practice, without exclusions and controlled condi-
tions [22].

To date, there are no meta-analytical reviews of psychological
interventions for PNES. This absence may be ascribed to the lack
of RCT’s, the customary design used for a meta-analytical review
and synthesis [23]. However, when considering the high social,
psychological and financial costs associated with PNES, there is an
imperative to utilize the current body of research to its full extent
[22]. Additionally, considering the complex nature of PNES,
combined with the difficulty and high cost of RCT’s, it is unlikely
there will be a sufficient number of RCT’s conducted in the near
future for this type of meta-analysis to be performed. Meanwhile,
uncontrolled and naturalistic treatment evaluations in clinical
service-settings provide valuable information in their own right
[22]. Increasingly, as in other areas of health research, the
combination of large amounts of observational literature and the
pressure for timely, accurate clinical information compels
researchers to utilize observational studies using meta-analysis
[21]. Combining this diffuse body of research will also enable
this information to be more readily accessible, and therefore,
help educate clinicians of current evidence-based treatments for
PNES [23].

The aim of this systematic review and meta-analysis is to
evaluate and examine the available evidence from the previous
20 years regarding the effectiveness of psychological interventions
in the management of PNES. Using meta-analysis, this study
predicts that psychological interventions for PNES will be shown to
be associated with both seizure freedom and reductions in seizure
frequency of 50% or more.

2. Method

2.1. Protocol

A review protocol for this study was developed in December
2015 and is available upon request from the author (PC).

2.2. Eligibility criteria

Eligible studies were required to be published electronically in
peer reviewed science journals in the English language between
1996 and 2016. PNES was defined as the experience of non-
epileptic seizures of psychological origin as diagnosed by a
neurologist, psychologist or psychiatrist and confirmed by
electroencephalogram (EEG) or video-EEG (vEEG). Given the
important differences between PNES in adults and children, the
studies required the inclusion of participants aged 16 years and
older (>50% of the participants are ≥16 years). Due to the limited
amount of research in this area, the search was open to all
prospective human studies, including controlled and before-after
non-controlled studies including case series. Qualitative single
case studies, and retrospective studies were excluded, as were
review articles and conference abstracts.

Studies were included if they evaluated the effectiveness of at
least one psychological intervention undertaken to lessen the
frequency of PNES. An intervention was considered to be
psychological in nature if it was based on a psychological theory
or model specifically designed to alter psychological processes
thought to underlie or significantly contribute to pain, distress, and
disability [24].

Additional selection criteria included using seizure frequency as
an outcome measure. Regardless of the method of reporting, this
criterion was included in the qualitative synthesis. Studies selected
as eligible for quantitative synthesis were required to provide
sufficient information on the primary outcome of seizure
frequency so as to enable the calculation of either seizure
reduction and/or seizure freedom rates. Studies were excluded if
they examined the effectiveness of non-psychological interven-
tions (i.e. medication) or focused on psychological interventions
that targeted other outcomes (i.e. employment status, cost
efficacy).

2.3. Search and selection strategy

In order to decide which studies to include in the analyses, an
extensive literature search was conducted utilising two online
academic databases, MEDLINE via OvidSP and PsychINFO (see
Appendix A). To do this, a search strategy was developed using a
wide-ranging pool of MeSH/thesaurus terms tailored to each
database (see electronic search strategy for MEDLINE via OvidSP
database in Table 1). The search was conducted by the author (PC)
and included records from 1996 to July 2016. If the article title
indicated relevance then the abstract was read. The complete
article was read if the abstract indicated the article met the
inclusion criteria. Following this, reference lists from selected
studies were examined for additional relevant papers. The authors
consulted in the event of any queries and discrepancies were
resolved by discussion. This search was conducted on 3 June
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