

Stressful life events and maltreatment in conversion (functional neurological) disorder: systematic review and meta-analysis of case-control studies



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Summary

Background Stressful life events and maltreatment have traditionally been considered crucial in the development of conversion (functional neurological) disorder, but the evidence underpinning this association is not clear. We aimed to assess the association between stressors and functional neurological disorder.

Methods We systematically reviewed controlled studies reporting stressors occurring in childhood or adulthood, such as stressful life events and maltreatment (including sexual, physical abuse, and emotional neglect) and functional neurological disorder. We did a meta-analysis, with assessments of methodology, sources of bias, and sensitivity analyses.

Findings 34 case-control studies, with 1405 patients, were eligible. Studies were of moderate-to-low quality. The frequency of childhood and adulthood stressors was increased in cases compared with controls. Odds ratios (OR) were higher for emotional neglect in childhood (49% for cases vs 20% for controls; OR 5.6, 95% CI 2.4–13.1) compared with sexual abuse (24% vs 10%; 3.3, 2.2–4.8) or physical abuse (30% vs 12%; 3.9, 2.2–7.2). An association with stressful life events preceding onset (OR 2.8, 95% CI 1.4–6.0) was stronger in studies with better methods (interviews; 4.3, 1.4–13.2). Heterogeneity was significant between studies (I^2 21.1–90.7%). 13 studies that specifically ascertained that the participants had not had either severe life events or any subtype of maltreatment all found a proportion of patients with functional neurological disorder reporting no stressor.

Interpretation Stressful life events and maltreatment are substantially more common in people with functional neurological disorder than in healthy controls and patient controls. Emotional neglect had a higher risk than traditionally emphasised sexual and physical abuse, but many cases report no stressors. This outcome supports changes to diagnostic criteria in DSM-5; stressors, although relevant to the cause in many patients, are not a core diagnostic feature. This result has implications for ICD-11.

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Introduction

Conversion (functional neurological symptom) disorder (DSM-5) refers to patients who have neurological symptoms in the absence of neurological disease, encompassing symptoms such as limb weakness, seizures, and movement disorders. Such disorders are one of the most common reasons for neurological referral (16% of new referrals)¹ and are as disabling and distressing as neurological counterparts such as multiple sclerosis or epilepsy.² Traditionally, the disorder has been diagnosed on both the absence of neurological disease and the “conflicts or other stressors [that] precede the initiation or exacerbation of the symptom or deficit”.³ However, the most recent edition of DSM-5 dropped the association with conflicts or other stressors as an explicit diagnostic criterion and emphasised the need to find positive clinical features such as Hoover’s sign in functional leg weakness or a sudden prolonged motionless unresponsive episode with eyes closed in dissociative (non-epileptic) seizure. This change has not been universally welcomed and whether ICD-11 will follow suit is uncertain.

Stressors, either recent life events, maltreatment around the time of symptom onset, or historical stressors, particularly childhood sexual abuse, have been considered key factors for the cause of functional neurological disorder since the time of Briquet’s 1859 Clinical and Therapeutic Treatise on Hysteria.⁴ In 1895, Breuer and Freud described the processes by which such psychological stress was converted into physical symptoms in their seminal Studies on Hysteria,⁵ giving the condition its name—conversion disorder—and a theory for cause that remains the bedrock of practice for most clinicians today.

However, critics of the conversion hypothesis have commented that the empirical evidence to support the hypothesis is poor and that the dominance of the theory distorts clinician’s appreciation of the limitations of the available literature and inhibits the development of alternate or expanded models.⁶

Previous reviews summarising studies of stressors, including maltreatment and stressful life events, in functional neurological disorder have either not been systematic,⁶ or have only reviewed non-epileptic

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Research in context

Evidence before this study

In the past decade, interest in conversion (functional neurological) disorders has upsurged. Largely dismissed during the latter part of the 20th century as a historical entity that was usually the product of misdiagnosis, high-quality evidence now shows that such disorders are common, disabling, and can be diagnosed accurately. The dominant view of cause had been that these symptoms arose as a consequence of the conversion of psychic distress into physical symptoms. With new research came new theories of cause; in particular, increasingly sophisticated models of mechanism based in neurosciences. These theories challenged the dominant view of psychological stressors being converted into physical symptoms. One view is that these new studies were complementary and simply explained the mechanism of conversion. Other clinicians and researchers took the stance that this mechanism was alternate and that conversion disorder could occur in the absence of identifiable exposure to stressors. DSM-5 took the latter view and was explicit that the presence of such stressors was no longer required, although paradoxically went for a compromise name: conversion (functional neurological symptoms) disorder. This opinion was not met, with universal approval, and passionate debate still exists, but has often been shaped by individuals citing case examples from their own practice and less attention has been paid to the existing data from case-control studies. As a group of clinical researchers who have been involved in this debate but from opposing perspectives, we sought to systematically review the available literature. We searched PubMed and Science Direct for case-control studies in English from 1965 to Nov 4, 2016, with the search terms (“psychogenic” OR “conversion disorder” OR “non-epileptic”) AND (“abuse” OR “life event”) AND (“control” OR “controlled” OR

“case-control”). This search was supplemented by reviewing the reference lists of eligible studies and previous reviews. After removing duplicates and ineligible studies, we included 34 studies in our systematic review and meta-analysis.

Added value of this study

To our knowledge, this review provides the most comprehensive aggregation of the evidence from case-control studies since 1965. We covered the full phenotype of conversion (functional neurological) disorder, including both seizure disorders and motor or sensory disorders. We examined the frequency of stressful life events and of different types of maltreatment in childhood and adult life in patients with conversion (functional neurological) disorder and in both healthy controls and controls with neurological disease and psychiatric disorders. In addition to a qualitative review of individual studies, we quantitatively evaluated the association between stressors and functional neurological disorder, and did subgroup and sensitivity analyses to investigate sources of bias to understand the limitations to our data.

Implications of all the available evidence

Our results show that the rate of childhood and adult stressful life events and maltreatment, particularly emotional neglect, is increased in patients with conversion (functional neurological) disorder compared with controls. The association was stronger in cases of childhood onset and when we compared with healthy controls as opposed to disease controls. However, a proportion of cases report no stressors. We concluded that stressors are relevant to the cause and development of conversion (functional neurological) disorder and therefore a potential treatment target, but exposure to such stressors is not an essential diagnostic feature. Our findings support the changes to DSM-5 and have implications for ICD-11.

seizures^{7,8} or childhood sexual abuse.⁷ These reviews suggested an association of stressors and functional neurological disorder, but were of limited scope. When looking at more broad phenotypes, reviews of somatic symptom disorders have notionally included functional neurological disorder, but either did not identify much of the existing primary literature⁹ or were focused on functional somatic syndromes such as irritable bowel syndrome or chronic fatigue, which overlap with, but are different from, functional neurological disorder.¹⁰

Technically, the study of maltreatment—used here as an umbrella term for sexual and physical abuse as well as emotional neglect—and stressful life events is challenging for many reasons. These challenges include patients’ willingness to disclose sensitive information (and possibly even awareness of it or of its potential relevance), recall bias, difficulty determining over what timeframe stressors are relevant, whether those that are present are relevant to cause, and the selection of appropriate controls. The use and selection of control

groups is of particular importance as the rates of recent and historical stressors vary in different clinical (whether psychiatric or neurological) and healthy populations.

Furthermore, the descriptive terminology is at times ill defined and, in doing a systematic review, we are partly dependent on the definitions used in individual studies. Thus, during the process of data amalgamation, it becomes inevitable that compromises are made between the uniqueness of an individual event and its psychological context, and the need to impose a taxonomy to allow quantitative study. We have developed a glossary of terminology that, although imperfect, allows for clarity and reproducibility (appendix).

We aimed to do a systematic review of the association between childhood and adult stressful life events and maltreatment and conversion (functional neurological) disorder by reviewing all quantitative case-control studies since 1965 and comparing frequencies in functional neurological disorder populations with those frequencies in healthy, neurological or psychiatric disorder control

See Online for appendix

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