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Review Article

Factitious disorder: a systematic review of 455 cases in the professional literature☆

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ABSTRACT

Objective: Patients with factitious disorder (FD) fabricate illness, injury or impairment for psychological reasons and, as a result, misapply medical resources. The demographic and clinical profile of these patients has yet to be described in a sufficiently large sample, which has prevented clinicians from adopting an evidence-based approach to FD. The present study aimed to address this issue through a systematic review of cases reported in the professional literature.

Method: A systematic search for case studies in the MEDLINE, Web of Science and EMBASE databases was conducted. A total of 4092 records were screened and 684 remaining papers were reviewed. A supplementary search was conducted via GoogleScholar, reference lists of eligible articles and key review papers. In total, 372 eligible studies yielded a sample of 455 cases. Information extracted included age, gender, reported occupation, comorbid psychopathology, presenting signs and symptoms, severity and factors leading to the diagnosis of FD.

Results: A total of 66.2% of patients in our sample were female. Mean age at presentation was 34.2 years. A healthcare or laboratory profession was reported most frequently ($N = 122$). A current or past diagnosis of depression was described more frequently than personality disorder in cases reporting psychiatric comorbidity (41.8% versus 16.5%) and more patients elected to self-induce illness or injury (58.7%) than simulate or falsely report it. Patients were most likely to present with endocrinological, cardiological and dermatological problems. Differences among specialties were observed on demographic factors, severity and factors leading to diagnosis of FD.

Conclusions: Based on the largest sample of patients with FD analyzed to date, our findings offer an important first step toward an evidence-based approach to the disorder. Future guidelines must be sensitive to differing methods used by specialists when diagnosing FD.

1. Introduction

Factitious disorder (FD) with physical symptoms is a psychiatric disorder in which sufferers intentionally fabricate illness, injury or impairment in order to gain hospital admission and undergo medical procedures, without any obvious gain [1]. It is considered to be one of the most challenging disorders in medical experience [2]. Patients with FD may exaggerate or lie about a medical condition, mimic or "act out" medical symptoms, interfere with diagnostic investigations or even directly self-induce illness or injury [3]. In contrast to malingerers, who fabricate medical need for reasons of clear external reward (such as evading military service or gaining disability benefits), the motivations of patients with FD are 'almost always obscure' [4] and may include a desire to receive affection and care, an "adrenaline rush" from undergoing medical procedures or a sense of control from deceiving healthcare professionals [5]. Patients with FD may expose themselves to a considerable risk of iatrogenic harm [6]. Indeed, one patient with FD described by Robertson and Hossain [7] admitted to having undergone 42 surgical procedures over the course of 850 admissions to 650 different hospitals. Fatality due to FD appears to be rare, but it does occur [8–11].

Studies on FD demonstrate the heavy impact of unnecessary investigations, treatments and hospital admissions on the healthcare system. Healthcare costs in individual cases of FD have exceeded $200,000 [12] and even $1,000,000 [13]. A patient with FD may also have a considerable psychological impact on hospital staff involved in their care. Staff may feel anger at having been "duped" by the patient and "cheated" of the time and support they have expended [14,15], or they may experience guilt for allowing themselves to be drawn into the emotional conflicts that frequently arise in cases of FD [16,17].

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Most doctors will encounter at least one patient with FD over the course of their clinical practice [18]. However, the exact prevalence of FD in hospital settings is currently unknown [19–21]. FD may account for between 0.6% and 3% of referrals from general medicine to psychiatry [22–24] and between 0.02% and 0.9% of cases reviewed in specialist clinics [25–28]. A recent study surveying physicians' own estimates of the presence of factitious symptoms among their patients reported a higher prevalence rate of 1.3% [29]. Rates of FD may be greatly increased in patient populations whose reported problems are diagnostically challenging [30,31] or have received significant public attention [32].

Although FD has been recognized by clinicians for centuries [33], if not millennia [34], the first extensive study on FD appears in Asher's initial description of "Munchausen's syndrome" in 1951. However, since that time, the term "Munchausen's syndrome" has become a source of confusion in both clinical practice and the published literature [3]. The correct usage of the term is to denote a particularly severe and chronic presentation of FD [33], but "Munchausen's" is often used interchangeably with "factitious". Other terms used for FD include "hospital hopper syndrome", "hospital hobo syndrome" and "thick chart syndrome", and they frequently display a level of irony — e.g. "black hole patients" or "peregrinating problem patients". These terms reflect that patients with FD can be derided by healthcare professionals.

Patients with FD may fabricate medical need in several ways. The variety of methods available to these patients is limited in principle only by their level of dedication, imagination and medical knowledge [35] but is dependent in practice upon the nature of the medical problem they intend to fabricate. For example, a patient with FD attempting to fabricate urological disease may falsely report the presence of chronic urinary discomfort, deliberately withhold urine to simulate acute anuria [36], add blood to urine samples to simulate hematuria [37] or actually induce a urinary tract infection by self-injection with bacterial cultures [38]. A patient attempting to fabricate a dermatological condition may be restricted to simulating a lesion (e.g. by discoloration of the skin with ink [39]) or creating an actual lesion through self-mutiliation [40] or other means [41]. Patients with FD may employ several of these methods at once [3] and frequently present with diverse symptomatology. The wealth of medical knowledge now available on the Internet may enable patients lacking a background in healthcare to present with complex medical problems. It is seldom possible to diagnose FD with conviction [3] but when the diagnosis is made, it usually follows an exhaustive series of medical procedures undertaken to rule out an organic explanation for the patient's problems.

Early detection of FD is thus paramount in order to limit wastage of healthcare resources and harm to patients. Early management of FD may also facilitate improved outcomes for patients with the disorder [3]. However, the clinical and demographic profile of patients with FD has not been clarified with a sufficiently large sample [33]. We consider such knowledge to be an important first step in the development of an evidence-based approach to the early detection and management of FD in clinical settings. The majority of the published literature on FD consists of case reports and series, which are a valuable source of information but may present a misleading clinical picture of the disorder in isolation [42]. Indeed, assumptions about the characteristics of patients with FD abound in the professional literature — one troubling example being the idea that the majority of patients with the disorder are male, as specified in the DSM-IV despite the clear lack of research supporting such a statement [43]. Although recommendations have been published concerning the detection of FD (e.g. see Ref. [33]), these recommendations have not been supported by broad evidence on how FD is diagnosed by clinicians on a wider scale or how methods for detecting medical deception may vary among medical specialties. Similarly, guidelines for management of FD (e.g. see Ref. [44]) have been written in the absence of substantial data concerning the severity of the methods typically adopted by patients with FD — or indeed the suicide risk and psychiatric comorbidity associated with the disorder. This is information integral to effective management of FD [17].

What is therefore needed is a comprehensive and systematic review of the case reports and series available in the professional literature, as has been undertaken previously with child and adolescent FD [45], FD imposed upon another or "Munchausen-by-proxy syndrome" [46–48] and other uncommon disorders [49–51]. Use of this method has enabled authors to examine the clinical and demographic characteristics of samples of patients larger than would be feasible for comparable empirical studies.

Unfortunately, only a limited number of reviews have been published on FD, and those published to date have been mainly limited to a small number of cases from single medical specialties — recently, cardiology [32], neurology [52], obstetrics and gynecology [53], ENT [54], oncology [55] and dermatology [56]. Authors who have aggregated cases across specialties have limited their sample to cases of FD that have been treated [57] or detected by laboratory testing [58-60], and they have therefore analyzed only a minority of cases available in the professional literature.

Thus, it was the aim of this study to undertake a comprehensive, systematic review of all cases of FD with physical symptoms published in the professional literature to date, to characterize for the first time the basic demographic and clinical profile of patients with FD in a large sample and to compare these features among medical specialties. This review was restricted to adult cases of FD, as a full review of child and adolescent FD was beyond the scope of this study and has previously been conducted [45].

2. Method

2.1. Types of study

A systematic search was conducted for all case studies and series that reported on adult patients eligible for a DSM-5 diagnosis of FD with primarily physical symptoms [1] on the basis of the clinical information provided by the author(s). This search included cases where the diagnosis of FD was described in other terms, such as 'dermatitis artefacta' and 'Munchausen's', or was classified according to a comparable diagnostic system, such as DSM-IV [43] or ICD-10 [4]. Chart reviews and larger case series were excluded if they did not also describe cases individually. Following the conservative methodology outlined by Kanaan and Wessely [52], studies were excluded if they reported cases in which no firm diagnosis of FD could be made.

2.2. Search strategy

A broad keyword search of literature published in English between January 1, 1965 and July 27, 2015 was conducted. MEDLINE, Web of Science and EMBASE databases were searched using the terms, factit*, munchausen*, artefacta* and artefactua*. Records with 'by proxy' or 'imposed upon another' were not automatically filtered out of the search results in order to ensure that case series reporting both FD and FD imposed upon another were included. A total of 4256 records were returned following exclusion of duplicate records, of which 4092 were retrieved for abstract review. A total of 748 records were identified as potentially eligible, of which 684 were retrieved for full-text review. A total of 333 studies were selected for inclusion after full-text review. The bibliographies of eligible studies were also screened, in addition to the bibliographies of multiple review papers [52–59] and the results of a GoogleScholar search utilizing terms identical to the keyword search. These supplementary search processes yielded a further 39 eligible studies. Search formulae for MEDLINE, Web of Science and EMBASE databases are provided in Section 1 of the supplemental material. The PRISMA (Preferred Reporting Items for Systematic Reviews and Meta-Analyses) flow chart for the search process is provided in Section 2 of the supplemental material.