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

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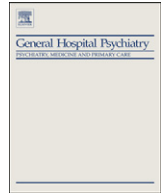**101** PUBLICATIONS   **1,327** CITATIONS

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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.
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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].

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-mutilation [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.

2.3. Data collected

A mean number of 9.1 new cases per year was reported over the review period, with a tendency toward higher values in more recent years: 1965–1975 (3.5/year), 1975–1985 (7.4/year), 1985–1995 (11.2/year), 1995–2005 (12/year) and 2005–2015 (11.3/year). Single cases were extracted from 86% of studies, while the remaining 14% contributed multiple patients.

The following quantitative and qualitative variables were obtained (percentage of data found indicated in parentheses) for each case: *age* (99%), *gender* (100%), *reported occupation* (47%), *index presentation of FD* (100%), *psychopathology* (37%) and *factors leading to diagnosis of FD* (100%). When recording *reported occupation*, patients were only coded as 'unemployed' when this was specified by authors. Similarly, a lack of mention of patient *psychopathology* was not interpreted as an absence of comorbid psychiatric symptomatology, which was only coded when authors clearly specified that a psychiatric assessment or chart review had taken place with nothing of significance found. Marital status, race and ethnicity and education were reported only in a small minority of cases and were therefore not addressed in this review.

Presentation of FD was extracted by recording the presenting sign(s), symptom(s) or diagnosis at admission. Each presentation was recorded as 'falsely reported', 'feigned' or 'induced' according to clinical information provided and categorized by medical specialty according to system affected and initial referral. Following Kanaan and Wessely [61], where a history of repeat presentations was described, initial presentation was taken to be the index presentation for the case.

Psychopathology was extracted by recording current or historic psychiatric diagnoses described by the author(s). Diagnoses were not recorded where there was significant doubt expressed by the author(s) concerning the veracity of psychiatric symptoms described.

Factors leading to diagnosis of FD were extracted using a checklist adapted from two surveys of clinical information that might raise suspicion of FD [33,44]. This checklist included 8 items outlined with examples in Table 1. Items on the checklist were coded only if the factor contributed to the diagnosis made by the author(s). Clinical information that did not contribute to the diagnosis made by the author(s), or contributed in retrospect only, was not assessed.

2.4. Analysis

IBM SPSS 23 [62] was used to calculate descriptive statistics. A narrative synthesis was undertaken to describe common presentations and fabrication methods reported by included studies.

3. Results

3.1. Demographics

Patients with FD were described worldwide: 249 from America (United States of America, 237; Canada, 8; Brazil, 2), 150 from Europe (United Kingdom, 94; Italy, 9; Germany, 8; Belgium, 7; Greece, 6; the Netherlands, 6; Poland, 4; Republic of Ireland, 3; Austria, 2; Croatia, 2; Denmark, 2; Spain, 2; France, 1; Hungary, 1; Macedonia, 1; Sweden, 1; Romania, 1), 5 from Africa (Tunisia, 2; Morocco, 1; South Africa, 1; Zimbabwe, 1), 42 from Asia (Japan, 13; Turkey, 13; India, 6; Saudi Arabia, 4; Israel, 3; Iran, 3), 7 from Australia, 1 from New Zealand and 1 from Cuba.

A total of 33.8% of patients with FD were male. Mean age at presentation was 34.2 years with a median of 32 years and a range of 61 (maximum, 79; minimum, 18). Table 2 contains a breakdown of age and gender by medical specialty. Patient occupation was described in 214 (47%) cases. In 122 of these cases, a healthcare/laboratory profession was reported. The single most common occupation described was nursing (N = 68),

Table 1
Factors leading to diagnosis of FD

#	Factor	Examples
1	**Past healthcare service use**	History of extensive healthcare service use; history of peregrination between healthcare services; history of FD confirmed by healthcare professional
2	**Information provided by patient**	Inconsistent, selective or misleading biographical information provided; evasive when history is taken; dramatic but unlikely medical history provided; unusual difficulty corroborating information provided; refusal to allow access to outside information sources
3	**Atypical presentation**	Symptoms predominantly occur when the patient is not under observation; course of illness is impossible, highly improbable or does not follow the natural history of the presumed diagnosis
4	**Unsubstantiated presentation**	Investigations normal or inconclusive
5	**Evidence of fabrication**	Physical evidence of fabrication discovered through search or surveillance; patient directly witnessed simulating disease
6	**Patient behavior**	Unusual medical knowledge or use of medical terminology; eagerness for medical procedures; aggression or defensiveness with healthcare staff; noncompliance with diagnostic or treatment recommendations; *pseudologia fantastica*; patient opposes psychiatric involvement while pursuing medical or surgical options
7	**Investigations indicating fabrication**	Investigations reveal mechanism of fabrication; investigations rule out organic etiology; evidence from investigations contradicts information provided by patient
8	**Treatment failure**	Appearance of new symptoms on commencement of treatment; symptoms worsen on commencement of treatment

3.2. Index presentation of FD

Table 3 contains a summary of presentations of FD by medical specialty. A narrative synthesis describing common presentations and fabrication methods reported by included studies is provided in Section 3 of the supplemental material. Across all specialties, 22.2% falsely reported disease/injury, 19.1% simulated disease/injury and 58.7% induced disease/injury. A full breakdown of FD severity by medical specialty is included in Table 4.

Table 2
Basic demographic characteristics of patients diagnosed with FD by medical specialty

Specialty	N	Age (SD; range [min; max])	% Female
Allergy and Immunology	8	27.9 (10.3; 31 [min 20; max 51])	88
Cardiology	44	38.4 (11.9; 47 [min 20; max 67])	23
Dermatology	43	40.6 (16; 61 [min 18; max 79])	79
Endocrinology	59	32.3 (9.4; 36 [min 18; max 54])	78
ENT	11	28.3 (10.9; 37 [min 19; max 56])	73
Gastroenterology	29	34.1 (11; 43 [min 19; max 62])	76
Hematology	27	34.4 (14.4; 53 [min 21; max 74])	74
HIV and Sexual Health	11	32 (12; 43 [min 19; max 62])	45
Microbiology and Infection	13	28.5 (6.3; 25 [min 18; max 43])	92
Neurology	32	34.4 (10; 46 [min 22; max 68])	44
Obstetrics and Gynecology	7	40.3 (16.1; 47 [min 21; max 68])	100
Oncology	12	31.6 (7.9; 26 [min 19; max 45])	92
Ophthalmology	18	32.1 (15.2; 55 [min 18; max 73])	83
Oral and Maxillofacial	8	27.4 (5.3; 15 [min 20; max 35])	75
Orthopedics and Trauma	34	30.1 (9.9; 38 [min 18; max 56])	53
Plastic and Reconstructive Surgery	13	33.4 (9.4; 31 [min 23; max 54])	77
Pulmonary and Respiratory	33	33 (12.8; 53 [min 19; max 72])	70
Rheumatology	9	36.9 (8.2; 25 [min 22; max 47])	67
Urology and Nephrology	30	34.9 (9.9; 35 [min 22; max 57])	53
Other	14	41.7 (8.5; 31 [min 22; max 53])	71

Table 3
Index presentations of cases by medical specialty

Specialty	N	Index presentations
Allergy and Immunology	8	Allergic emergency induced ($N=3$), signs of immune deficiency induced ($N=3$), allergic emergency simulated ($N=2$)
Cardiology	44	Retrosternal chest pain reported ($N=29$), hypertension induced ($N=5$), arrhythmia induced ($N=2$), unconsciousness and bradycardia induced ($N=1$), history of myocardial infarction reported ($N=1$), syncopal episodes simulated ($N=3$), syncopal episodes induced ($N=1$), history of syncopal episodes reported ($N=1$), ventricular tachycardia and myocardial infarction induced ($N=1$)
Dermatology	43	Generalized lesions induced ($N=10$), breast lesions induced ($N=7$), facial lesions induced ($N=4$), leg lesions induced ($N=3$), pyoderma gangrenosum simulated ($N=3$), pyoderma gangrenosum induced ($N=1$), hand lesions induced ($N=2$), genital lesions induced ($N=2$), purpura of the knee induced ($N=2$), subcutaneous nodules and abscesses induced ($N=2$), ulceration of elbow and forearm induced ($N=1$), acute erythematous eruption induced ($N=2$), neck lesions induced ($N=1$), arm lesions induced ($N=1$), generalized purulent lesions with lichenification induced ($N=1$), swelling of the hand induced ($N=1$), painful lobules of lower body induced ($N=1$), cheilorrhagia and cheilitis of lip induced ($N=1$)
Endocrinology	59	Recurrent hypoglycemia induced ($N=31$), signs and symptoms of Cushing's syndrome induced ($N=9$), thyrotoxicosis induced ($N=8$), diabetic ketoacidosis induced ($N=4$), history of diabetes reported ($N=2$), hyperglycemia induced ($N=1$), pheochromocytoma simulated ($N=1$), endocrine neoplasia simulated ($N=1$), symptoms and family history of multiple endocrine neoplasia reported ($N=1$), history of Zollinger-Ellison syndrome reported ($N=1$)
ENT	11	Facial swelling induced ($N=4$), airway distress reported ($N=2$), bleeding from mouth, nose, ears and eyes simulated ($N=1$), bleeding from ears simulated ($N=1$), airway distress feigned ($N=1$), cheilitis of lip induced ($N=1$), ear drainage simulated ($N=1$)
Gastroenterology	29	Severe diarrhea induced ($N=6$), diarrhea simulated ($N=2$), recurrent vomiting induced ($N=1$), hematemesis simulated ($N=5$), hematemesis and hematochezia reported ($N=1$), hematemesis induced ($N=1$), gastrointestinal bleeding induced ($N=3$), epigastric pain induced ($N=3$), rectal bleeding simulated ($N=1$), gastrointestinal bleeding simulated ($N=1$), deterioration of Crohn's disease induced ($N=2$), uriointestinal fistulae simulated ($N=1$)
Hematology	27	Anemia induced ($N=9$), purpura induced ($N=4$), hypercalcemia induced ($N=2$), hypokalemia induced ($N=1$), systemic mastocytosis reported ($N=1$), acute lymphoblastic leukemia reported ($N=1$), chronic myeloid leukemia reported ($N=1$), hemophilia reported ($N=1$), epistaxis induced ($N=1$), abnormal coagulation induced ($N=1$), signs of deep vein thrombosis simulated ($N=2$), sickle cell disease reported ($N=3$)
HIV and Sexual Health	11	History of HIV reported ($N=5$), history of AIDS reported ($N=3$), history of HIV-related Kaposi's sarcoma reported ($N=1$), history of AIDS-related disease reported ($N=1$), history of venereal disease reported ($N=1$)
Microbiology and Infection	13	Sepsis induced ($N=7$), septic arthritis induced ($N=3$), necrotizing fasciitis simulated ($N=1$)
Neurology	32	Chronic pain reported ($N=7$), paralysis or weakness simulated ($N=4$), unconsciousness simulated ($N=4$), cyclic hypersomnia simulated ($N=1$), seizures simulated ($N=3$), torsion dystonia simulated ($N=2$), hemifacial spasm simulated ($N=1$), symptoms of acute meningitis reported ($N=2$), migraine reported ($N=1$), scalp abrasions induced ($N=2$), signs of baroreflex failure induced ($N=1$), sciatica and urinary incontinence reported ($N=1$), blindness simulated ($N=1$), aphasia simulated ($N=1$), deterioration of Parkinson's induced ($N=1$)
Obstetrics and Gynecology	7	Vaginal bleeding induced ($N=5$), menorrhagia induced ($N=1$), vaginal discharge simulated ($N=1$)
Oncology	12	History of breast cancer reported ($N=4$), family history of breast cancer reported ($N=2$), ovarian cancer reported ($N=1$), cancer of small intestine reported ($N=1$), uterine cancer reported ($N=1$), Hodgkin's disease reported ($N=1$), adenocarcinoma of urinary bladder reported ($N=1$), symptoms of osteogenic carcinoma reported ($N=1$)
Ophthalmology	18	Keratoconjunctivitis induced ($N=6$), corneal damage induced ($N=4$), anterior scleritis induced ($N=2$), diplopia reported ($N=2$), acute endophthalmitis induced ($N=1$), eyelid swelling induced ($N=1$), crystalline keratopathy induced ($N=1$), signs of basal cell carcinoma induced ($N=1$)
Oral and Maxillofacial	8	Swelling of mandibular region induced ($N=2$), abrasion of oral mucosa induced ($N=2$), gingival ulceration induced ($N=1$), progressive facial pain reported ($N=1$), subluxation of jaw simulated ($N=1$)
Orthopedics and Trauma	34	Subcutaneous emphysema induced ($N=5$), chronic wound deterioration induced ($N=5$), pain induced ($N=5$), severe trauma simulated ($N=4$), severe trauma induced ($N=3$), subfascial emphysema induced ($N=2$), joint dislocation simulated ($N=2$), joint dislocation induced ($N=1$), chronic edema induced ($N=1$), burns induced ($N=1$), pyoderma gangrenosum induced ($N=1$), trauma reported ($N=1$), thigh abscess induced ($N=1$), suprapubic ulceration and vesicocutaneous fistula induced ($N=1$)
Plastic and Reconstructive Surgery	13	Wound deterioration induced following surgery ($N=9$), skin ulceration induced ($N=3$), deep muscular abscess induced ($N=1$)
Pulmonary and Respiratory	33	Asthmatic episodes simulated ($N=8$), acute respiratory distress simulated ($N=3$), hemoptysis simulated ($N=4$), hemoptysis reported ($N=3$), cystic fibrosis reported ($N=2$), pleuritic chest pain reported ($N=2$), intractable bronchorrhea reported ($N=1$), severe leg pain and pulmonary history reported ($N=1$), asphyxia simulated ($N=1$), inability to be weaned from ventilator reported ($N=1$), pneumothorax induced ($N=2$), hypoxemia induced ($N=1$), signs of collagen vascular disorder induced ($N=1$), inhalational pulmonary talcosis induced ($N=1$)
Rheumatology	9	Lobular panniculitis induced ($N=2$), nodular panniculitis induced ($N=1$), non-specific panniculitis induced ($N=2$), polyarthralgia and subcutaneous masses induced ($N=1$), systemic lupus erythematosus reported ($N=2$), arthritis simulated ($N=1$),
Urology and Nephrology	30	Severe renal pain reported ($N=13$), UTI induced ($N=5$), hematuria simulated ($N=3$), proteinuria simulated ($N=1$),

3.3. Psychopathology

Presence or history of comorbid psychiatric disorders was assessed in 170 patients. The most common comorbid psychiatric disorder found in this subsample was depression, which was identified in 41.8% of these patients. Other common disorders that were identified included personality disorder (16.5%), substance abuse (15.3%), anxiety (14.7%), functional neurological symptoms (5.3%) and eating disorders (4.1%). A total of 14.1% of patients reported current suicidal ideation or a history of suicide attempt(s). Authors reported the absence of comorbid psychopathology in 17.1% of the 170 cases.

3.4. Factors leading to diagnosis of FD

In the majority of cases (78%), an *unsubstantiated presentation* contributed to a diagnosis of FD. *Past healthcare service use* contributed to diagnosis in 47% of cases, *atypical presentation* in 40% of cases, *treatment failure* in 35% of cases, *investigations indicating fabrication* in 33% of cases, *patient behavior* in 31% of cases, *evidence of fabrication* in 31% of cases and *information provided by patient* in 22% of cases. In two cases, the diagnosis was made solely on the basis of a spontaneous confession. A full breakdown of these factors by medical specialty may be found in Table 5.

A spreadsheet providing a basic description of all studies included in this review is provided in Section 4 of the supplemental material.

4. Discussion

4.1. Demographic characteristics

The remarkable proportion of patients in our sample reporting an occupation related to healthcare or the laboratory (57%) supports

Table 4
FD severity by medical specialty

Specialty	N	False report of disease/injury	Feigned disease/injury	Induced disease/injury
Allergy and Immunology	8	0%	25%	75%
Cardiology	44	61%	16%	23%
Dermatology	43	0%	0%	100%
Endocrinology	59	7%	2%	92%
ENT	11	9%	45%	45%
Gastroenterology	29	10%	17%	72%
Hematology	27	26%	0%	74%
HIV and Sexual Health	11	100%	0%	0%
Microbiology and Infection	13	0%	0%	100%
Neurology	32	31%	56%	13%
Obstetrics and Gynecology	7	0%	29%	71%
Oncology	12	92%	0%	8%
Ophthalmology	18	6%	6%	89%
Oral and Maxillofacial	8	38%	0%	63%
Orthopedics and Trauma	34	3%	15%	82%
Plastic and Reconstructive Surgery	13	0%	0%	100%
Pulmonary and Respiratory	33	27%	58%	15%
Rheumatology	9	11%	22%	67%
Urology and Nephrology	30	27%	50%	23%
Other	14	29%	36%	36%
Median		**10.5%**	**15.5%**	**69%**
IQR		**29.25**	**34.25**	**64.25**

previous observations that the majority of patients with FD claim to have worked in these settings [21,63–68]. Krahn et al.'s [42] chart review of 93 FD patients found that a similar proportion (44%) of patients worked in a healthcare field. Of the 122 patients who made such a claim in our review, 114 were female, supporting Krahn et al.'s [42] identification of a subtype of FD consisting of female healthcare professionals. Overrepresentation of healthcare professionals in our sample may be due to publication bias, which will be discussed later. Alternatively, this result may be explained by the appeal of healthcare-related professions (in particular, nursing) for these patients. A career within a healthcare service may carry a similar appeal to deceiving clinicians for individuals predisposed to FD [69]. The "rush" of undergoing medical procedures reported in FD [5] may comparable to the "rush" of delivering them in a professional setting. On balance, our findings support the broad recommendation that healthcare professionals should be particularly vigilant for FD in patients who appear to have unusual medical knowledge or claim to have worked in related occupations [70].

The mean age of our sample at presentation (34.2 years) corroborates the results of several case series that were not included in our review [42,71–74]. This finding supports the understanding held by many authors that patients with FD present to healthcare services in early adult life [11,20,67,75–77]. An exception was noted with patients presenting with dermatological problems: they tended to be older (40.6 years) and included geriatric cases (Table 2). The finding of a female majority (66.2%) in our sample may conclude an ongoing confusion in the professional literature concerning the gender distribution of patients with FD. Despite several key case series [42,71,74] and reviews [52,56,57,78] indicating the opposite, numerous authors have made reference to a male predominance in cases of FD [54,79] — likely a result of the inclusion of a statement to this effect in DSM-IV [43] that was subsequently reversed in DSM-IV-TR [80] and abandoned in DSM-5 [1]. The case studies included in this review that were published before the release of the DSM-IV in 1994 (N = 164) did not support such a statement: 58.1% of patients in this subsample were female. Our total sample confirms that, overall, patients with FD tend to be female. However, important gender differences were observed among specialties (Table 2). For example, patients presenting with HIV-related, sexual health or neurological problems were predominantly men, and fewer than 25% of cardiac patients were female.

4.2. Severity of FD

Our review provided an unprecedented opportunity to compare the different methods employed by patients with FD in a large sample. We found that 58.7% of patients elected to induce illness or injury in themselves instead of attempting only to simulate (19.1%) or falsely report (22.2%) a medical problem. This preference would suggest that what has previously been regarded as an "extreme" variant of FD may in fact be its most common presentation. Patients with FD must be considered at significant risk of self-injury. This risk should be factored into any management plan for individuals suspected of medical deception. Friends, partners and family members should, if possible, be involved in this process in order to monitor the patient's access to tools (e.g. surgical instruments) and substances (e.g. prescription drugs, poisons) that may be used to induce injury or illness. Involuntary detention may even be indicated when patients with FD are socially isolated and using methods of illness induction that are difficult to control. Abuse of insulin (to induce hypoglycemia) and self-venesection (to induce anemia) are two such methods that were utilized by a disturbingly high number of

Table 5
Factors leading to diagnosis of FD by medical specialty

Specialty	N	Past healthcare service use	Information provided by patient	Atypical presentation	Unsubstantiated presentation	Evidence of fabrication	Patient behavior	Investigations indicating fabrication	Treatment failure
Allergy and Immunology	8	25%	0%	50%	63%	50%	38%	38%	50%
Cardiology	44	68%	41%	27%	91%	23%	50%	21%	32%
Dermatology	43	23%	5%	70%	79%	26%	16%	30%	42%
Endocrinology	59	29%	12%	31%	78%	32%	10%	73%	36%
ENT	11	73%	27%	82%	91%	55%	73%	18%	27%
Gastroenterology	29	38%	14%	35%	86%	41%	24%	52%	28%
Hematology	27	70%	22%	30%	78%	41%	22%	41%	19%
HIV and Sexual Health	11	82%	46%	18%	91%	9%	46%	0%	18%
Microbiology and Infection	13	54%	8%	15%	54%	69%	23%	62%	54%
Neurology	32	59%	28%	66%	84%	13%	63%	9%	31%
Obstetrics and Gynecology	7	57%	0%	29%	71%	29%	14%	43%	43%
Oncology	12	67%	75%	25%	50%	17%	25%	0%	0%
Ophthalmology	18	44%	6%	50%	67%	28%	44%	28%	33%
Oral and Maxillofacial	8	50%	0%	38%	75%	63%	25%	0%	100%
Orthopedics and Trauma	34	44%	35%	35%	56%	24%	27%	32%	32%
Plastic and Reconstructive Surgery	13	31%	15%	46%	69%	23%	31%	39%	77%
Pulmonary and Respiratory	33	55%	24%	46%	88%	27%	27%	15%	49%
Rheumatology	9	44%	22%	67%	89%	56%	33%	11%	44%
Urology and Nephrology	30	33%	20%	10%	90%	43%	23%	23%	17%
Other	14	50%	36%	43%	64%	7%	43%	36%	29%
Median		**50**	**21**	**36.5**	**78**	**28.5**	**27**	**29**	**32.5**
IQR		**30.75**	**26.75**	**22.5**	**24**	**25.25**	**20.75**	**28.5**	**20.5**

patients in our sample and led to fatality in a number of cases. Patients with FD who adopt these methods may present themselves as medically knowledgeable, but it is unlikely that they are fully aware of the probable adverse consequences of their behaviors.

Authors have assumed that patients with FD are intelligent and resourceful [16,63,66,70] and that these qualities are required to successfully deceive experienced clinicians [35]. However, these qualities are not required by patients with FD who induce illness or injury. Self-induction of illness or injury enables patients to reliably command attention from hospital staff [3] and may reflect a need to self-harm [5].

4.3. Psychopathology

Comorbid psychiatric disorders (or their absence) were described in only 37% of cases ($N = 170$). This finding may reflect the fact that the majority of the cases included in this review were not written by psychiatrists. Nonetheless, some observations may be made. Only 14.1% of patients were described as suicidal or as having a history of suicide attempts. It has been assumed previously that FD entails significant suicide risk and suicide attempts have been described (see Ref. [11] for a useful review). In this regard, our findings may provide a degree of reassurance. Similarly, no psychotic symptomatology was described in our sample to confirm an earlier hypothesis that FD is a defense against psychosis [81,82]. A surprising finding was the absence of personality disorders in all but a small minority of this subsample. The claim that FD is strongly associated with personality disorders (in particular, borderline personality disorder) is widespread in the professional literature [11,20,21,65,83–85] and has been included in multiple review articles [33,44]. Even so, the comorbid diagnosis reported most commonly in our sample was depression, providing support instead for an association between FD and mood disturbance [44,86]. However, the relationship between these two diagnoses is not clear. FD may be truly comorbid with depression due to shared risk factors for the two disorders, which include childhood abuse or neglect [87,88], parental failures [89–92], marital difficulties [93], substance abuse [94–96] and stressful life events [97,98]. Alternatively, FD may be secondary to depression — for example, as an expression of low self-esteem or a manifestation of the urge to self-harm, which has been linked to depressive symptoms [99,100]. In this case, it is plausible that treatment of depressive symptoms in cases of FD may lead to a reduction of factitious illness behavior.

4.4. Presentation of FD

Considerable variation was observed in the number of cases included in this review for each medical specialty. This variation may be explained by differential interest in FD among authors working in different specialties [52]. For example, the high number of dermatology cases eligible for this review may be the result of dermatologists' increased awareness of or interest in FD rather than a genuine "preference" of patients with FD. The inclusion of 'dermatitis artefacta' in the ICD predates the inclusion of FD by several decades [101,102]. Alternatively, variation among specialties may be explained by the relative difficulty of identifying FD within various medical specialties. For example, the preponderance of factitious hypoglycemia in this review may signal the comparative ease with which insulin abuse can be detected in the laboratory [58,59]. However, assuming that the distribution of cases across medical specialties corresponds to some degree to preferences of patients with FD, our findings demonstrate the need for health professionals working in endocrinology, cardiology and dermatology services to be specially watchful for FD.

As expected, patients with FD gravitated toward signs and symptoms leading to protocol-driven or "fast-track" admission, such as retrosternal chest pain. Similarly, patients made good use of widely available agents to induce serious illness, such as insulin, anticoagulants or thyroid hormones. This may have contributed to the high number of patients presenting with dermatological and endocrinological

problems. It is clear that patients with FD are capable of using their medical knowledge not only to simulate illness convincingly but also to find the "path of least resistance" to admission. Nevertheless, many patients were attracted to specialties with more complex disorders and a greater likelihood of discovery, such as cardiology and neurology. Kanaan and Wessely (2005) discussed this problem in their own review of neurological cases of FD, suggesting that the increased difficulty of simulating certain medical problems (due to modern imaging and laboratory investigations) may in FD be counterbalanced by increased 'reward' in the form of greater attention and sympathy. This suggestion would explain the relative popularity of oncology and cardiology to patients in our sample despite the difficulty often involved in fabricating cancer or coronary disease.

However, we might suggest that it is simply the case that patients with FD are able to make as much use of modern technology to maintain their deception as clinicians are to detect it, and for this reason, it is not as difficult for these patients to fabricate complex medical problems as we might assume. Numerous authors included in this review discuss the ease with which their patients were able to use the Internet to research their presentation of choice [103,104], forge medical reports or referral letters [105,106] and even purchase prescription medications [107]. The Internet may therefore enable patients with FD to be sufficiently versatile and adaptive in their deception to present to more challenging medical specialists [108]. In any case, our findings challenge the notion that the problems presented by FD will be overcome with innovations in health technology.

4.5. Factors leading to diagnosis of FD

As may be expected, a presentation unsubstantiated by objective clinical evidence was found to have facilitated discovery of FD in the majority of cases included in this review across all specialties. Our review therefore supports existing guidelines that caution health professionals to consider FD early when encountering patients whose complaints appear unsupported by physical examination, the results of investigations and so forth [33,44]. However, the variance among medical specialties that we observed in clinicians' use of other sources of information diagnose FD is unaccounted for by these guidelines and contrasts sharply with several claims made in the published literature. For example, it has been assumed that dermatologists are able to diagnose FD primarily on the basis of physical examination because skin lesions produced by patients with FD are morphologically atypical [109–111] and located on easily accessible areas of the body [112]. While this clinical picture facilitated diagnosis in 70% of the dermatological cases of FD that we reviewed, in a significant minority of cases, the appearance of lesions did not alert dermatologists to the possibility that they were being deceived. Similarly, although it has been argued that patients presenting with factitious endocrinological illness may be recognized by their surgical history [113], in the majority of cases involving such patients, such a history was either not described or not regarded as suspicious by endocrinologists. Future guidelines for the detection of FD in clinical settings must be data driven and sensitive to the practical realities of diagnosing the disorder in different medical specialties. This approach will help to avoid problematic assumptions of the kind discussed above and to offset the limitations of "one size fits all" clinical guidelines for FD. Our findings, accrued from the largest sample of patients with FD analyzed to date and a broad range of medical specialties, provide a robust starting point for such an approach.

4.6. Limitations

A number of limitations to this review must be acknowledged. Firstly, although case reports constitute the best source of knowledge currently available about FD, the sample used in this review is nonrandom and unlikely to be fully representative. Studies appeared more likely to be published if they presented an unusual manifestation

of FD, a novel technique for detecting FD or an entertaining account. A culture of "one-upmanship" may therefore have deterred potential authors from submitting cases of FD that were less severe or similar to previously published cases. Publication bias of this kind may also have accounted for the overrepresentation of healthcare professionals in our sample. Healthcare professionals are clearly capable of using their expertise to fabricate medical need more convincingly, as is shown in several reports included in this review [64,114,115]. It therefore stands to reason that cases involving healthcare professionals would be overrepresented in a literature biased toward ingenious patients. This publication bias may be compared to the 'file-drawer problem' described in metaanalysis [116], although the preponderance of less severe and nonnovel cases of FD included in our sample provides reassurance.

Secondly, it is plausible that our sample contains duplicates, as patients with FD are typically treated by several clinicians in the course of their deception — potentially across several regions [117] or countries [118] — any of whom may publish the case [21]. Cases explicitly describing a previously reported patient were excluded, but publication of cases in low-impact journals with a limited readership may have restricted the extent to which authors could be aware that they were describing the same patient as a previous case report.

Thirdly, because we extracted only basic demographic and clinical information from our sample, we could not examine the relationship between the results of this review and patient outcomes — nor could we relate our data to the etiology of FD. Eastwood and Bisson [57] conducted a similar systematic review of cases ($N = 316$) in order to evaluate management techniques for FD. However, inconsistent reporting of outcomes in case studies on FD significantly limited the extent to which they were able to use their analysis to make recommendations for the treatment of the disorder. This would suggest that original research is required to address these research questions assertively.

Finally, although care was taken to include only cases meeting a highly conservative interpretation of the DSM-5 criteria for FD [42], it possible that factors integral to the diagnosis of FD (e.g. absence of external reward) were not considered by authors, as many healthcare professionals do not encounter FD in everyday clinical practice and are unfamiliar with its diagnostic criteria [119]. Indeed, the most convincing patients with FD do not appear to meet diagnostic criteria for the disorder at all. Cases of FD reported in the professional literature (and consequently, in this review) may represent only the accounts of the patients least capable of avoiding detection or the clinicians most capable of detecting them.

5. Conclusion

FD is one of the most challenging disorders in medical experience but the clinical and demographic profile of the disorder has yet to be clarified with a sufficiently large sample. Accordingly, we conducted a systematic review of 455 cases of FD in the professional literature, the largest sample analyzed to date. Our findings provide several clinical recommendations (see below) and a strong first step toward an evidence-based approach to detection and treatment of FD.

5.1. Clinical recommendations

- Clinicians should be particularly vigilant for FD in patients who are female, in early adult life and claiming to have worked in healthcare or a laboratory
- Although patients with FD may appear in any specialist setting, endocrinology, cardiology and dermatology services should expect to encounter more
- FD is associated with low suicide risk, but these patients typically self-induce illness or injury and should therefore be considered at high risk of permanent damage, if not fatality
- FD is associated with depressive symptoms more than personality disorders and may be improved by treatments for depression

Appendix A. Supplementary data

Supplementary data to this article can be found online at http://dx.doi.org/10.1016/j.genhosppsych.2016.05.002.

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