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Management of Factitious Disorders: A Systematic Review

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Key Words
Factitious disorder · Munchausen's syndrome · Global Improvement Scale

Abstract
Background: The literature regarding the management of factitious disorder (FD) is diverse and generally of case reports or case series. To date there has been no systematic review of the effectiveness of management techniques. ***Methods:*** Systematic review of all evidence reporting the management and subsequent outcome in FD. Data were extracted and outcomes were assessed using an adaptation of the Global Improvement Scale. Results were analysed by parametric statistical tests; a meta-analysis was not possible. ***Results:*** Thirty-two case reports and 13 case series were eligible for inclusion. Analysis of the case reports found no significant difference in outcomes between confrontational and non-confrontational approaches [$t(29) = 0.72$, $p = 0.48$], between treatment with psychotherapy compared to no psychotherapy [$t(30) = 0.69$, $p = 0.48$], and when psychiatric medication had been prescribed compared with not [$t(30) = 0.35$, $p = 0.73$]. A trend was observed that a longer length of treatment lead to better outcomes, but this was not significant [$F(5, 26) = 1.17$, $p = 0.35$]. The consecutive case series demonstrated that many FD sufferers were not engaged in treatment and were lost to follow-up but did not provide any strong evidence regarding the effectiveness of different management approaches. ***Conclusions:*** There is an absence of sufficient robust research to determine the effectiveness of any management technique for FD. The establishment of a central reporting register to facilitate the development of evidence-based guidelines is recommended.

Introduction

Factitious disorder (FD), although a psychiatric condition, challenges practitioners in most fields of medicine. Categorised as an Axis I DSM-IV condition, it is diagnosed when there is intentional production or feigning of physical or psychological signs or symptoms where the incentive is to assume the sick role and external incentives for the behaviour are absent [1]. In the ICD-10 (F68.1) it is defined as repeated and consistent feigning of symptoms with obscure motivation for the behaviour and best interpreted as a disorder of illness behaviour and the sick role [2]. Once an organic cause has been excluded, the main differential diagnoses are malingering where external incentives are present for the intentional behaviour, and somatoform disorders in which both unconscious symptom production and unconscious motivations are present. Distinction between these disorders can be very

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difficult and the diagnosis of FD can only be wholly confirmed if observation of symptom-producing behaviour occurs or it is admitted. As neither of these scenarios is frequent, diagnosis usually remains only a high index of suspicion [3].

A community study of 2,363 people in Italy found the lifetime prevalence of FD to be 0.1% [4]. Estimated prevalence rates of FD have varied in other studies from 0.6% of 15,000 psychiatric consultations [5] to 9.3% of referrals to the National Institute for Allergy and Infectious Disease with fever of unknown origin lasting over 1 year [6]. It is generally acknowledged that the disorder is underreported and constitutes a real problem to medical services [7]. Munchausen's syndrome [8] is an extreme form of FD and is estimated to account for around only 10% of the factitious population [9], despite its overrepresentation in the literature. In Munchausen's by proxy the feigning of symptoms is in another person for the purpose of assuming the sick role by proxy and will not be considered further in this paper. The phenomenon of autodestructive behaviour has also attracted interest, encompasses a broader spectrum of disorders than FD alone and reported prevalence rates range from 0.032 to 9.36% [10].

The literature regarding management of FD is scarce and of poor quality. Many different techniques have been reported, primarily focussing on confrontational [11–13] versus non-confrontational [14] approaches. Various forms of psychological, medical, surgical and conservative treatment methods have also been reported. Several publications have provided useful reviews of the management of FD [15, 16] but to date there has been no systematic review of the management of FD or any clear evidence to suggest that one treatment method is beneficial above and beyond others.

Given the limited existing literature, we decided to perform a systematic review of published evidence regarding the management of FD and then use this evidence base to hypothesise an effective management strategy which can be implemented in clinical practice and tested in future research.

Method

Data Sources

A systematic bibliographic search was undertaken to identify all evidence regarding the treatment and management of FD from databases (EMBASE, Medline, PsycINFO and CINAHL), and the Cochrane Library with each database being searched from inception to December 2005. A search string of keywords was used; 'factitious disorder' or 'Munchausen's syndrome' and 'treatment' or 'management' or 'therapy' or 'psychotherapy' or 'pharmacotherapy'. Additional papers were found by hand searching the references of retrieved articles. The search was restricted to papers with English language abstracts.

Study Selection

All systematic reviews, randomised controlled trials, controlled trials, case series or case reports which appeared in peer-reviewed journals were eligible for inclusion. To be included, studies had to be of FD or Munchausen's syndrome that had been confirmed or was considered the most likely diagnosis in patients of either gender aged over 16 years; cases of Munchausen's by proxy were excluded. Papers were only included if they described the management techniques used and subsequent outcomes. The searching and selection were done by the authors. Any disagreements with regard to inclusion or exclusion of a study were resolved by discussion.

Quality Assessment

All papers deemed eligible for assessment were assessed for quality using a standardised form. This form incorporated factors used in systematic reviews within psychiatry [17] but was specifically designed for this study. The criteria included clarity of the description of techniques used, evidence of selection bias (e.g. selected versus consecutive selected cases), whether follow-up was reported for all patients in the series, and whether there was independent assessment of outcome using a standardised scale.

Data Extraction

All included papers were scrutinised and relevant data were extracted using a specially designed form. The data were extracted in the categories of demographic information, details of the FD, the method of diagnosis, the occurrence and form of any confrontation, therapies used, the duration of therapies and the patient condition at the last reported follow-up. The accuracy of data extraction was double checked by the second author.

Assessment of Outcomes

As a result of the absence of the use of standardised outcome measures, the authors objectively assessed the studies using an adaptation of the Global Improvement Scale (GIS) from the Clinical Global Impression scale [18] to quantify the change in overall condition of reported patients. The papers were reviewed and scored by both authors, there was good concordance between the scores of each assessor and any disagreements were resolved by discussion.

Data Synthesis

An initial narrative synthesis was undertaken to describe the characteristics, quality and outcomes of the studies. There were insufficient trials of good quality to perform a true meta-analysis of the results. SPSS 12 was used for statistical analysis of extracted data and GIS scores. Case reports and case series were analysed separately due to their heterogeneity. For the case report data parametric statistical testing was used to compare the mean differences in outcomes across different variables. Independent t tests (two tailed) were used to analyse the mean difference in outcome variables with two conditions (e.g. confrontation and no confrontation) and one-way ANOVAs were used to analyse the

mean difference in outcomes across variables with more than two conditions, all tests were applied at a 5% level of significance. The distribution of the data for the sample size of 32 was close to normal and therefore the sample size of 30 or more and normal distribution preconditions were satisfied for the use of parametric tests. The data from the case series could not be synthesised in this way.

Results

Trial Flow

The search identified 132 papers for potential inclusion in the review, no systematic reviews, randomised controlled trials or controlled trials were identified. A total of 45 papers were eligible for inclusion in the review; 32 case reports and 13 case series. The main reasons for exclusion of papers were poor reporting of management strategies and outcomes or that they were anecdotal case histories focussed on presentation and diagnosis. Some large case reviews were excluded because there was insufficient detail to determine which treatment methods gave more successful outcomes than others [13].

Case Reports

Study Characteristics

Table 1 includes the characteristics of the case reports. There were 8 males and 24 females with an age range of 19–64 years (mean = 32.0, SD = 10.7). Seventeen patients had been diagnosed with Munchausen's syndrome, of these 5 were male. Duration of factitious illness ranged from 2 months to 40 years (mean = 6.5 years, SD = 7.5). Psychiatric comorbidity was common; 50% had another Axis I diagnosis and 40% had an Axis II diagnosis.

Quality Assessment

Case reports are flawed in terms of proving the efficacy of an intervention, in particular because of the risk of positive reporting bias and the absence of any form of control. There was considerable variation in the amount of information provided regarding demographic details of patients and the management techniques used. Only 10 (31.3%) reports made reference to diagnostic criteria. Only 6 papers (18.8%) reported follow-up at 12 months or more post-discharge from treatment. No papers reported an independent assessment of outcome or the use of a standardised assessment scale.

Management

Confrontation

Seventeen (53.1%) patients were confronted with their diagnosis, 14 (43.8%) were not and confrontation was not reported once (3.1%). Of those who were confronted, 14 had a non-punitive approach (82.4%), two had an accusative approach (11.8%) and the exact method was not reported in one (5.9%).

Different non-punitive confrontational techniques were used. In some, the patient was presented with diagnostic test results and it was explained that as they did not match the clinical picture it was believed that symptoms were being feigned [27]. Another technique was to tell the patient that whilst doctors believed the symptoms were being produced by the patient treatment was available [26, 28–30]. One accusative confrontation occurred in an angry, non-therapeutic atmosphere; the authors concluded that this was critical in the subsequent success of treatment [31]. One patient was repeatedly and vigorously confronted with the diagnosis whilst in a locked psychiatric ward from which she was unable to escape [12].

A variety of scenarios were reported in those cases where confrontation did not take place. In some cases confrontation was not required as the patient had admitted to fabricating illness [33–36]. Some authors reported that confrontation was not used for fear of damaging the therapeutic relationship [37, 38]. For one patient, a version of inexact interpretation was adopted in which there was free access to a hospital bed for a year and the patient was in control of when they came to hospital but a response to treatment was expected [19]. Jamieson et al. [39] reported a technique where 2 psychiatrists collaborated; one acted as a helper and one as a confronter and through alternate interactions with the patient, enabled her to acknowledge the diagnosis.

Other Interventions

Nineteen patients (59.4%) received only one type of treatment, the others (40.6%) received multiple therapies as outlined in table 1. Psychotherapy was the only form of therapy in 16 patients (50.0%) but in many the exact regimen was not fully described or techniques had been combined. Six (18.8%) cases reported the concurrent delivery of psychotherapy and medication. Other reported combinations were psychiatric interventions with physiotherapy and occupational therapy [24, 29], and surgical treatment of a wound fistula followed by psychotherapy and both antidepressant and antipsychotic medication [47]. One case reported monitoring of the patient but no specific therapy was mentioned [28]. Two cases reported

Table 1. Case report characteristics and outcomes

Authors	Confrontation technique	Single/ multiple therapies	Treatment description	Treatment location	Treatment length	GIS score (0–7)
Spiro, 1968 [35]	No confrontation (self-admission)	Single	Psychotherapy (not described)	Inpatient then outpatient	6 months	5
Fras and Coughlin, 1971 [44]	Repeated non-punitive confrontation	Multiple	Psychodynamic psychotherapy + anti-psychotic + social worker involvement	Mostly outpatient	2 years	2
Stone, 1977 [12]	Accusative confrontation	Multiple	Psychotherapy + antidepressant	Inpatient then outpatient	10 months	2
Yassa, 1978 [41]	Not reported	Single	Dynamic behavioural approach and supportive psychotherapy	Inpatient	3 years	2
Tucker et al., 1979 [31]	Carefully planned non-punitive confrontation.	Single	Intensive psychotherapy	Inpatient	11 months	2
Jamieson et al., 1979 [39]	Non-punitive confrontation	Single	Behavioural psychotherapy	Outpatient	4 months	3
Serafin et al., 1983 [47]	Accidental confrontation, with unconscious motivation emphasised	Multiple	Supportive psychotherapy + antidepressants + antipsychotics and surgery to repair fistula	Inpatient	2 months	3
Klonoff et al., 1983 [22]	No confrontation (previous attempts unsuccessful)	Multiple	Behavioural therapy delivered by 2 psychologists + biofeedback and relaxation techniques	Outpatient	15 months	2
Mayo and Haggerty, 1984 [34]	No confrontation (self-admission)	Single	Psychoanalytic psychotherapy	Outpatient	16 months	4
Batshaw et al. 1985 [33]	No confrontation (previous attempts unsuccessful)	Single	Insight-oriented psychotherapy and behaviour modification	Inpatient then outpatient	12 months	2
Gordon and Chrys, 1985 [25]	Non-punitive confrontation with supporting evidence	Single	Psychotherapy and stopping of medications	Inpatient	5 weeks	4
Earle and Folks, 1986 [42]	Non-punitive confrontation by physician	Multiple	Cognitive/supportive psychotherapy + antidepressant	Inpatient then nursing home	17 months	2
Kallen et al., 1986 [40]	No confrontation; aimed to educate patient through alternative method	Single	Symptomatic treatment focussed on speech articulation and relaxation techniques	Inpatient then outpatient	5 months	3
Simmons et al., 1987 [64]	No confrontation	Single	Behavioural therapy	Inpatient	2 months	4
Johnson et al., 1987 [30]	Non-punitive confrontation	Single	Supportive psychotherapy	Outpatient	6 months	2
Schoenfeld et al., 1987 [32]	Angry, accusative confrontation by psychiatrist	Single	Psychotherapy focussed on life coping-skills	Outpatient	4 years	1
Savard et al., 1988 [49]	Non-punitive confrontation (had been confronted previously)	Single	Psychotherapy (initially refused)	Outpatient	2 months	7
Harrington et al., 1988 [28]	Non-punitive confrontation	Single	Monitoring of patient only	Outpatient	12 months	3
Schlesinger et al., 1989 [23]	Non-punitive confrontation	Single	Psychotherapy and behaviour modification	Inpatient	5 months	3
Higgins, 1990 [48]	No confrontation, establishment of alliance with GP	Multiple	Supportive authoritarian approach adopted by GP + antidepressants and antipsychotics	Outpatient (GP co-ordinated)	8 years	1
Christensen and Szlabowicz, 1991 [27]	Non-punitive confrontation with diagnostic evidence	Single	Psychotherapy + withdrawal from antipsychotics.	Outpatient	2 months	2

Table 1 (continued)

Authors	Confrontation technique	Single/ multiple therapies	Treatment description	Treatment location	Treatment length	GIS score (0–7)
Parker, 1993 [36]	No confrontation (self-admission)	Single	Psychotherapy	Outpatient	8 months	3
Schwarz et al., 1993 [19]	Inexact interpretation; previous confrontation unsuccessful	Multiple	Behavioural psychotherapy + antidepressant + open access to hospital bed	Patient choice	12 months	2
Arya, 1993 [45]	No confrontation	Multiple	Psychiatric treatment (not specified) + analgesia for pain	Inpatient	2 years	2
Guziec et al., 1994 [26]	Non-punitive confrontation	Single	Supportive and insight-oriented psychotherapy	Outpatient	12 months	3
Feldman and Duval, 1997 [24]	Non-punitive confrontation with family present	Multiple	Psychological counselling + physiotherapy + OT	Outpatient	5 months	2
Kwan et al., 1997 [29]	Non-punitive confrontation	Multiple	CBT + physiotherapy + OT	Inpatient	6 weeks	4
Leonardou et al., 2002 [37]	No confrontation for fear of damaging therapeutic relationship	Single	Psychoanalytic psychotherapy and mileu therapy	Inpatient	2 months	1
Hirayama et al., 2003 [21]	Non-punitive confrontation	Multiple	Psychotherapy + antipsychotic and oral iron	Outpatient	15 months	7
Kubota et al., 2003 [38]	No confrontation for fear of damaging therapeutic relationship	Single	Levothyroxine as day-patient to treat feigned hypothyroidism	Outpatient	3 years	2
Yanik et al., 2004 [43]	No confrontation	Multiple	Psychiatric consultations + antidepressant	Outpatient	2 months	2
Oh et al., 2005 [46]	No confrontation	Multiple	Antidepressant + treatment for panniculitis	Inpatient	2 months	3

long-term, multi-faceted innovative approaches to treatment. In one, the patient was given open access to a hospital bed for a year whilst receiving behavioural therapy and antidepressants [19]. In the other, a GP took a consistent supportive yet authoritarian approach to a patient, who was also prescribed antidepressant and antipsychotic medication [48].

Quantitative Data Synthesis

The frequencies of GIS scores across the reviewed papers are included in table 1. The mean GIS score was 2.81 (SD 1.45) suggesting a small improvement at follow-up. Only seven (21.9%) had a score of 4 or higher representing no change or a worsening of condition. Two (6.3%) were assigned a value of 7; in both of these cases, the patient died [18, 48] through a successful suicide. Three had a GIS score of 1 [32, 37, 49] but no common themes emerged from these cases.

Confrontation

Analysis of the difference in mean GIS between patients who had been confronted (mean = 3.0, SD = 1.6) and not confronted (mean = 2.6, SD = 1.2) (reported for n = 31) gave a non-significant result [$t(29) = 0.72$, $p = 0.48$].

Other Interventions

A single categorisation system for the different treatments was difficult due to the heterogeneity of the treatments described. Therefore, different aspects of the treatments have been analysed. An independent samples t test (two tailed) comparing single or multiple treatments gave a non-significant difference in mean GIS (single treatment mean = 2.8, SD = 1.4; multiple treatments mean = 2.8, SD = 1.5) $t(30) = 0.14$, $p = 0.89$. Analysis comparing the difference in mean outcome between the interventions which incorporated psychotherapy (n = 27, mean = 2.9, SD = 1.5) and interventions with no psychotherapy (n = 5, mean = 2.4, SD = 0.9) gave a non-significant result, $t(30) = 0.69$, $p = 0.48$.

An independent samples t test comparing mean difference in GIS outcome following treatment where psychiatric medication had been prescribed (n = 9, mean = 2.7, SD = 1.7) compared with those where it had not (n = 23, mean = 2.9, SD = 1.4) gave a non-significant result, $t(30) = 0.35$, $p = 0.73$.

There was a trend of greater improvement with longer treatment (<2 months mean = 3.0, SD = 1.8; 2–6 months mean = 3.0, SD = 1.0; 6–12 months mean = 2.6, SD = 0.6; 12–18 months mean = 3.8, SD = 2.4; 18–24 months mean = 2.0, >24 months mean = 1.5, SD = 0.6), but this was not significant [$F(5, 26) = 1.17$, $p = 0.35$]. The mean difference in GIS between treatment as an inpatient (n = 15, mean = 2.9, SD = 1.4) or as an outpatient (n = 16, mean = 2.8, SD = 1.5) was non-significant [$t(29) = 0.34$, $p = 0.73$].

Case Series

Study Characteristics

The characteristics of the 13 case series are presented in table 2, they varied in size from 2 to 93 patients. A total of 284 patients were reported; 193 (68.0%) were female, 67 (23.6%) were male and in 24 (8.4%) gender was not documented. Patient ages ranged from 14 to 70 years (mean = 33.7, SD = 7.6). The duration of factitious illness was only reported for 218 patients, the mean for these was 6.31 (SD = 9.4) years. Most papers did not distinguish between FD and Munchausen's syndrome; only 7 (2.5%) patients were reported as having a diagnosis of Munchausen's syndrome. Seven papers (53.8%) made reference to co-morbid psychiatric diagnoses; from those papers (195 patients), 113 patients (57.9%) were reported as having co-morbid psychiatric disorders (Axis I or Axis II). Other papers referred to co-morbidities with depression, schizophrenia and personality disorders.

Quality Assessment

There was much variation in the quality of reporting. Only six (46.2%) described a consecutive series of patients [9, 50–54], the other seven (53.8%) described a group of selected patients which had been treated by the authors. Five (38.5%) referred to diagnostic criteria. Patient demographics and histories, the management techniques used and the duration of follow-up were reported with wide variation in detail. Follow-up information was only reported for 112 (39.4%) patients. The duration of follow-up was only reported in 9 papers (56 patients) and ranged from 1 month to 15 years (mean = 45 months, SD = 22.9). Seven (53.8%) did not follow-up all patients in the series, which may confound the evidence that they present. Two (15.4%) reported outcomes as assessed by another clinician [52, 55], and one paper used a standardised scale (the Malan scale) to assess the change in patient condition [56].

Management

Confrontation

Confrontation techniques were not always reported and were generally poorly described but included non-punitive approaches [51, 58, 59], a mixture of confrontational and non-confrontational strategies [53, 60], and a double-binded face-saving technique [54, 61].

Other Interventions

Table 2 details the wide range of treatments reported across the case series with some papers describing different techniques in different groups of patients. It was not always clear how the outcomes related to individual treatments across the case series [52]. The larger case series demonstrated that only a small proportion of patients who were identified with FD engaged in treatment [9, 53]. Many reported techniques were multidisciplinary in nature [56, 61]. Plassmann [56] described a long-term psychotherapeutic approach which had its foundations in the treatment of borderline personality disorder; patients initially received inpatient clinical psychotherapy followed by outpatient psychotherapy which had lasted up to 5 years at the time of reporting.

Quantitative Data Synthesis

A range of outcomes was seen, the GIS scores are included in table 2. The median value for the GIS was 0 (not reported) as the majority of patients (60.6%) were not followed up. Four patients (3.6%) were reported deceased. In the 7 papers where follow-up was reported for over 80% of patients (a total of 40 patients), the median GIS was 2 (improvement). Further analysis was not possible due to the wide variation in reporting between the case series.

Discussion

Principal Findings

There is an absence of robust research to determine the effectiveness of any management technique for FD. The best level of evidence was level III [62] indicating an absence of randomised controlled trials or controlled trials. The consecutive case series demonstrated that many FD sufferers are not engaged in treatment and are lost to follow-up.

Table 2. Case series characteristics and outcomes

Authors	Cases	Series selection	Confrontation technique	Treatment descriptions	Outcome descriptions	GIS score[1]
O'Shea and McGennis, 1982 [57]	2	Patients	Not reported	Informal psychotherapy with behavioural approach focussed on social skills	Improvement (n = 2)	(2–3)
Maurice-Williams and Marsh, 1985 [52]	14	Cases of simulated paralysis, 10 years neurosurgical admissions.	Conveyed that nature of behaviour was understood	Amyteal abreaction (n = 6), hypnosis (n = 2), psychotherapy (n = 1) pharmacotherapy (n = 2) placebo and reassurance (n = 7)	Improvement (n = 2), reoccurrence of symptoms (n = 3), no follow-up (n = 9)	0 (0–5)
Grunberger et al., 1988 [51]	10	Cases of factitious hypoglycaemia in 13 year period	Non-punitive with persistence until patient acknowledgement	Not described in detail, multidisciplinary approach	Dead (n = 2), much improved (n = 3), minimal improvement (n = 3), lost to follow-up (n = 1)	3 (0–7)
Solyom and Solyom, 1990 [55]	2	Patients	None; informed that they would be treated accordingly	CBT and physiotherapy (faradic massage)	Good improvement (n = 2)	(1–3)
Sutherland and Rodin, 1990 [50]	10	Cases of FD referred to consultation-liaison service, 3 years	9 confronted; exact technique unknown	Outpatient psychotherapy (NOS) (n = 2)	No reoccurrence of symptoms (n = 2), death (n = 1), no follow-up (n = 7)	0 (0–7)
Spivak et al., 1994 [58]	2	Patients	Non-punitive	Psychotherapy focussing on emotions and experiences (n = 2), ECT (n = 1), antipsychotic medication (n = 1)	Physical improvement, still psychosocial problems (n = 2)	3 (3–3)
Teasell and Shapiro, 1994 [61]	3	Patients	Double-binded, face-saving interpretation	Strategic-behavioural intervention involving psychotherapy, physiotherapy, OT and nursing with specific goals for patients	Rapid good improvement (n = 2), partial improvement (n = 1)	2 (1–3)
Plassmann, 1994 [56]	24	Patients	Not described; flexible, non-aggressive approach	Inpatient clinical psychotherapy then long-term psychotherapy	Variable improvement (n = 12), refusal of therapy (n = 12)	0 (0–4)
Freyberger et al., 1994 [53]	70	Hospitalised FD patients, 9 years seen by psychosomatic consultants	Accusative (n = 13), indirect (n = 58), no confrontation (n = 12)	Psychodynamic psychotherapy inpatient/outpatient (n = 17)	Improvement (n = 9), no change/worse (n = 3), continuing treatment (n = 1)	0 (0–4)
Shapiro and Teasell, 1997 [54]	24	Consecutively admitted patients rehab ward, 9 years	Double-binded, face-saving interpretation	Strategic-behavioural intervention (physiotherapy); psychological counselling in some patients	Complete improvement (n = 15), significant improvement (n = 3), minimal improvement (n = 2)	0 (0–4)
Al-Qattan, 2001 [60]	28	All patients FD upper limb, 6 years	Various techniques; confrontational and non-confrontational	Psychotherapy, pharmacotherapy or no treatment beyond confrontation but numbers not reported	Successful treatment (not qualified) (n = 11)	0 (0–5)
de Fontaine et al., 2001 [59]	2	Patients	Non-punitive, by surgeon and psychiatrist	Plastic surgery (n = 2); additionally psychotherapy (n = 1), monitoring in pain clinic (n = 1)	Wounds healed (n = 2)	(2–4)
Krahn et al., 2003 [9]	93	Computer list, FD diagnosis, 20 years	Confrontation (technique not specified) (n = 71); psychiatric consultation (n = 80)	Psychiatric treatment (n = 19), ongoing care (n = 29)	Ongoing care (n = 29), known deceased (n = 2) unknown (n = 62)	0 (0–7)

[1] Values are given as median (range).

Table 3. FD management recommendations

a	A comprehensive psychiatric assessment should be completed to identify comorbid psychiatric illnesses, and suicide risk
b	One person should have primary therapeutic responsibility
c	The multidisciplinary team should be involved with all being aware of the psychiatric assessment, risk assessment and treatment plan
d	If confrontation takes place, this should be non-punitive and supportive in nature
e	A treatment plan should be individualised to the patient
f	Comorbid illnesses should be treated appropriately
g	Long-term therapy or support should be provided to aid recovery and transition back into a 'normal' life

Strengths and Weaknesses of the Study

This review was conducted systematically using comprehensive searches with predetermined inclusion and exclusion criteria, and standardised assessment tools. Only limited quantitative data synthesis was possible. A formal meta-analysis could not be performed, but this study has provided what is likely to be an accurate picture of the current evidence base and should form the basis for the development of a strategy to improve knowledge in this area. Despite the absence of evidence for effectiveness of the management techniques considered, the risk of bias towards positive outcomes is very high with authors being more likely to present their successes than their failures and a similar risk of publication bias. The very variable lengths of follow-up also compromise interpretation. The paucity of good research in this area is likely to have been contributed to by FD being a relatively rare and difficult to detect condition that is notoriously difficult to manage with engagement often being a major problem [63].

Clinical Implications

Although no definite conclusions can be made from these data, it has been possible to make some observations regarding effective treatment methods. It seems likely that longer-term management plans are required for many FD sufferers and that a key challenge is how to achieve good engagement with therapy. Consistency of care and the adoption of a holistic approach to management may be fundamental in achieving effective outcomes. The absence of an observed difference between inpatient and outpatient management suggests that outpatient management will usually be more appropriate for reasons of cost effectiveness. The main role of pharmacological therapy in FD seems likely to be in treatment of co-morbid psychiatric disorder [42, 48]. The reported successes of both confrontational [32] and non-confrontational approaches [37, 48] along with treatments that focused on the presenting symptoms [38, 49] suggest that various strategies may be helpful but do not really help the clinician to select a particular management plan.

In two of the four case reports where a negative outcome was documented, suicide had occurred. Many authors have highlighted the unfavourable prognosis and risk of suicide in patients with FD [65]. Clinicians should remain vigilant to this and routinely assess suicide risk in suspected FD sufferers. The results of this review do not allow for the development of an evidence-based management pathway for FD but do allow some tentative management recommendations (table 3) to be made that require further evaluation.

Future Research

There remain many unanswered questions. In order to improve the current evidence base, we believe that a network of clinicians with an interest in this area should be established, a central reporting register developed and standardised patient data routinely collected and analysed. Such systems are already in place for other rare conditions such as Huntington's disease [66] and CJD [67] with the emergence of better quality research than could have been produced without their development. Rothwell [68] recently highlighted the general neglect of good observational research within medical academia leading to a failing in effective clinical practice. This is true for FD and if conducted would form a foundation for the phased development and testing of complex interventions that have the potential to be effective for FD in the future.

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