FACTITIOUS DISORDERS OF THE UPPER LIMB IN SAUDI ARABIA

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This prospective study investigated 28 patients with factitious disorders of the upper limb. Several cases did not fit any of the well-known factitious syndromes such as SHAFT or Münchhausen syndrome. Patients were divided into five groups, three of which comprised active mutilators who either induced wounds, introduced foreign bodies or induced arm swelling and oedema. Passive mutilators were considered in a separate group and were defined as those who complained of factitious pain or numbness of the limb, for which there was no secondary gain regarding employment or compensation: these patients underwent surgery and hence persuaded the surgeon to do the mutilation. The fifth group of patients exhibited hand posturing and were divided into two subgroups: patients with psychological hand posturing and those seeking secondary gain (malingerers). The clinical presentation, diagnosis and outcome are described for each group.
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Factitious disorders of the upper limb occur in a variety of forms and there are several classification systems (Table 1). SHAFT syndrome (''sad, hostile, anxious, frustrating and tenacious'') results in passive mutilation, as the patient complains of various symptoms and tries to persuade the hand surgeon to ''do the cutting'' (Graham et al., 1999; Kasdan et al., 1998). In contrast, Münchhausen's syndrome describes active, though usually hidden, self-mutilation. Patients with Münchhausen's syndrome seek medical treatment for factitious illness and invent fanciful and elaborate historical details to confound and intrigue the hand surgeon. The motives are quite variable and include a desire to fool the omniscient ''father image'' doctor and to gain attention. The term Munchausen's syndrome refers to the 18th-century figure Baron von Münchhausen who invented dramatic whimsical tales which won him legendary fame. Virtually all medical references misspell ''Münchhausen'' as ''Munchausen'', and thus the medical syndrome is commonly spelled with one ''h'' (Smith, 1975). The best-defined form of factitious oedema of the upper limb is Secretan's syndrome. Romm (1986) reviewed the history of Henri François Secretan, a Swiss physician who examined workers for insurance companies and described a specific condition which results in peritendinous fibrosis of the dorsum of the hand (Whitney and Jones, 1995). Patients with Secretan's syndrome are also called ''wall-bangers'' because they strike the dorsum of their hands until they produce a brawny subcutaneous oedema. Finally, patients with conversion phenomena assume abnormal hand postures, such as a clenched fist, stiff index finger, and dysfunctional finger postures (Grunert et al., 1991). The most common posture is rigid flexion of the proximal interphalangeal joints of the ulnar three fingers with free use of the index and thumb, the so-called clenched fist syndrome or psycho-flexed hand (Frykman et al., 1983). The diagnosis is confirmed by the demonstration of normal nerve conduction and electromyogram studies, and occasionally by examination under anaesthesia.

Grunert et al. (1991) classified factitious disorders of the upper limb into 3 groups: wound manipulators (which will include some patients with SHAFT and all patients with Münchhausen's syndrome); factitious oedema (which includes patients with Secretan's syndrome); and hand deformity (which includes patients with clenched fist syndrome). Kasdan et al. (1998) prefer to classify psychological disorders of the upper limb according to motivation and intentional sign and symptom production. Malingerers have conscious motivation and intentionally produce signs and symptoms, whereas patients with factitious and psychological disorders have unconscious motivation. However, intentional sign and symptom production is present in factitious, but not in psychological, disorders.

The aim of this prospective study was to investigate the pattern of factitious disorders of the upper limb in Saudi Arabia, to demonstrate that many cases do not fit into any of the existing syndromes described in the literature, and finally to offer an extended classification for these disorders.

MATERIALS AND METHODS

All patients with factitious disorders of the upper limb who were seen by the author during the last 6 years were included in this prospective study. The following data were documented: age, sex, marital status, type of factitious disorder, occupation, social or job-related problems and previous psychiatric illness. Time off work and the payment of compensation were noted. Finally, diagnosis, treatment and outcome were documented.

RESULTS

A total of 28 patients with factitious disorders of the upper limb were included. Their ages ranged from 16 to 60 (mean, 33) years. There were 19 women and nine men. Only three were married and living with their

Table 1—Three classification systems for factitious disorders of the upper limb

Common syndromes	*Grunert et al. (1991)*	*Kasdan et al. (1998)*
1. SHAFT syndrome	1. Wound manipulation	1. Malingering
2. Münchhausen's syndrome	2. Factitious oedema	2. Factitious disorders
3. Secretan's syndrome	3. Hand deformity	3. Somatoform disorders
4. Clenched fist syndrome		

Table 2—The classification of the 28 patients with factitious disorders of the upper limb

Group/Subgroup	*Number of patients*
I. Wound inducers	
a. By inducing infection	3
b. By burns	2
c. By other means	3
II. Foreign-body introducers	
a. Air	1
b. Organic foreign body	1
c. Non-organic foreign bodies (such as needles)	4
III. Oedema inducers	
a. Secretan's dorsal hand oedema	0
b. Other forms of oedema (such as using a tourniquet)	3
IV. Passive mutilators	1
V. Hand posturing	
a. Psychological disease	3
b. Job-or compensation-related malingering	7

partners: the others were single, divorced, separated or widows/widowers. Patients were divided into five groups according to the type of factitious disorder as shown in Table 2.

Wound inducers

All eight patients in this group were single or divorced women and the wounds were induced by infection, burning the skin with a hot metal object, scratching or other unknown mechanisms. Interestingly, infections were always induced with a syringe and needle. One patient with recurrent wounds exhibited characteristics of the SHAFT personality (sad and hostile and requesting surgery) and another had the Münchhausen's personality (inventing different stories and requesting medical treatment and dressings). The personality and presentation of the other six patients were not typical for either the SHAFT or Münchhausen's syndrome. An example is shown in the following case report.

Case report

A 20-year-old girl, who was intelligent and had a pleasant personality, presented with recurrent infection and a chronic wound on the dorsum of the left hand which had caused extension contractures of the fingers (Fig 1). She had been treated with regular dressings for 4 years and had previously undergone six split-thickness skin graft procedures which had all failed because of infection. The patient was admitted to hospital and underwent psychiatric evaluation and daily dressings, and the hand was put in a bulky dressing to prevent manipulation. The wound fully healed in 3 weeks and the patient was then discharged home. Two weeks later, she presented to the clinic requesting release of the extension contractures of the fingers. After discussing the case with the psychiatrist, it was decided to delay any reconstructive surgery for at least 6 months, and the patient was informed of that decision. During the next 6 months, she kept her psychiatric appointments and developed no new wounds or infections. She then underwent release and grafting of the contractures. On the 2nd post-operative day, a nurse caught her injecting the wound through the dressing with a needle and syringe. A severe hand infection resulted which responded to broad-spectrum antibiotics, but the graft was lost. The patient did not come back for further follow-up.

Fig 1 A 20-year-old girl with a self-inflicted recurrent infection and a chronic wound on the dorsum of the left hand.

Foreign body inducers

There were six women in this group. The most common foreign body was all, or part of, a sewing needle that had

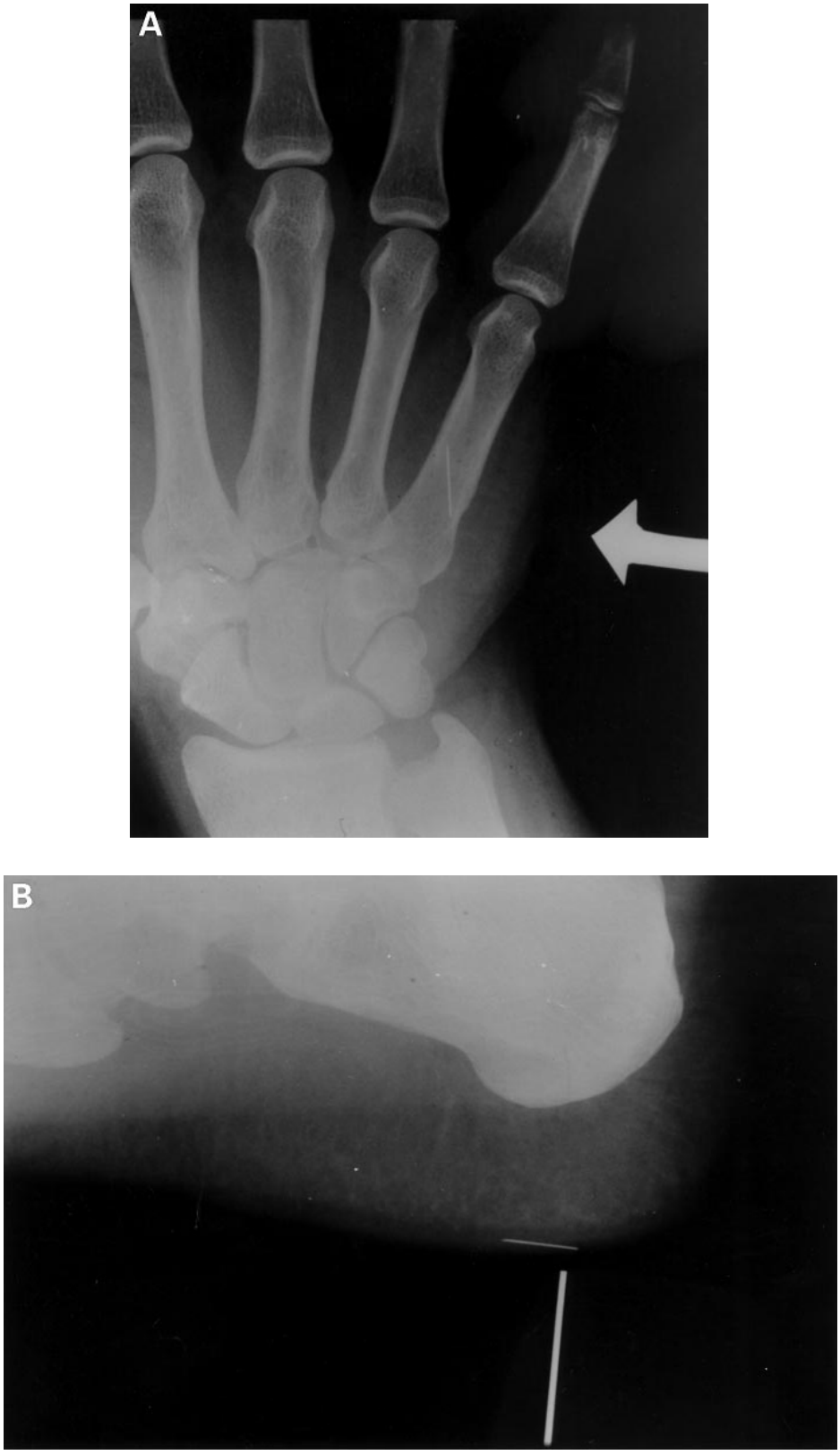

Fig 2 A 25-year-old girl with a sewing needle in her hand. (A) Radiograph of the hand; (B) on reviewing the medical charts, an old radiograph was found showing a sewing needle in the foot. On asking the family, it was discovered that she had undergone removal of sewing needles from various parts of her body on eight occasions during the previous 4 years. These procedures were performed at four different hospitals.

been introduced superficially under the skin of the hand or forearm. All patients claimed that needle introduction was accidental. The diagnosis of intentional needle introduction was relatively easy to make after reviewing the medical charts (Fig 2). One patient presented several times having introduced air into the forearm using a syringe and a needle (Fig 3). The most difficult case to diagnose was one of organic foreign body introduction.

Case report

A 50-year-old widow presented with multiple subcutaneous masses in the left antecubital fossa and adjacent forearm (Fig 4A). The patient denied any past history of trauma or a medical illness, such as tuberculosis. A biopsy was performed and the excised subcutaneous mass was sent for histology and mycobacterial and fungal cultures. Histology revealed multiple granulomas without caseation (Fig 4B) and staining was negative for acid-fast bacilli and fungi. All the cultures were negative. The patient was referred to the medical clinic at which she underwent extensive investigations for various granulomatous diseases and chronic inflammatory conditions, all of which were negative. The possibility of self-inflicted injury was raised and on reviewing her past medical history we found that she had previously been admitted to a psychiatric ward for depression and had been caught once injecting her arm with vegetable matter. The patient was referred to a psychiatrist and never returned for follow-up.

Oedema inducers

No cases of Secretan's oedema were seen. However three single young girls (aged 16–18 years) induced upper limb oedema by applying a tourniquet around the upperarm or forearm. These were referred from various general practitioners as cases of lymphoedema and the clinical diagnosis of self-inflicted injury was made by the observation of a sharp demarcation line in the skin (Fig 5). A psychiatric consultation resulted in a cure within one month of treatment in all cases.

Passive mutilators

Only one case of passive mutilator was seen in the series.

Case report

A 40-year-old housewife presented with a long history of unilateral non-specific hand pain. She had previously seen several surgeons requesting surgery. Various investigations were performed, including radiographs of the cervical spine, thoracic outlet and the hand. Nerve conduction studies were normal, but she eventually underwent a carpal tunnel release which did not improve her symptoms. The nerve conduction studies were repeated on presentation to the author, and were negative. Her history was inconsistent at her follow-up visits and it became apparent that her pain had started after family problems. Treatment for depression by the psychiatrist resulted in complete relief of pain.

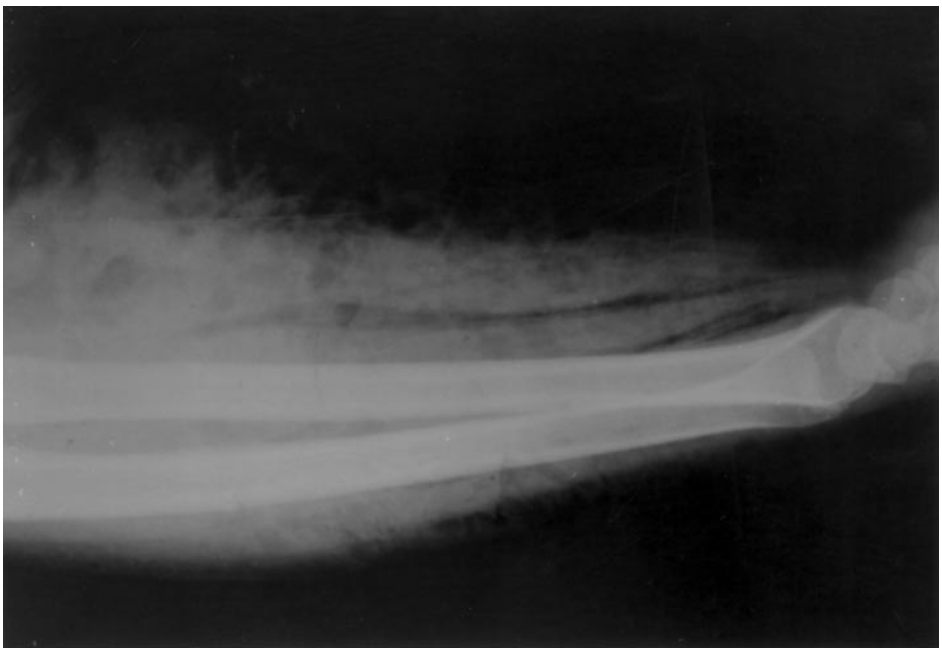

Fig 3 The radiographic appearance of the air introduced into the forearm using a needle and a syringe.

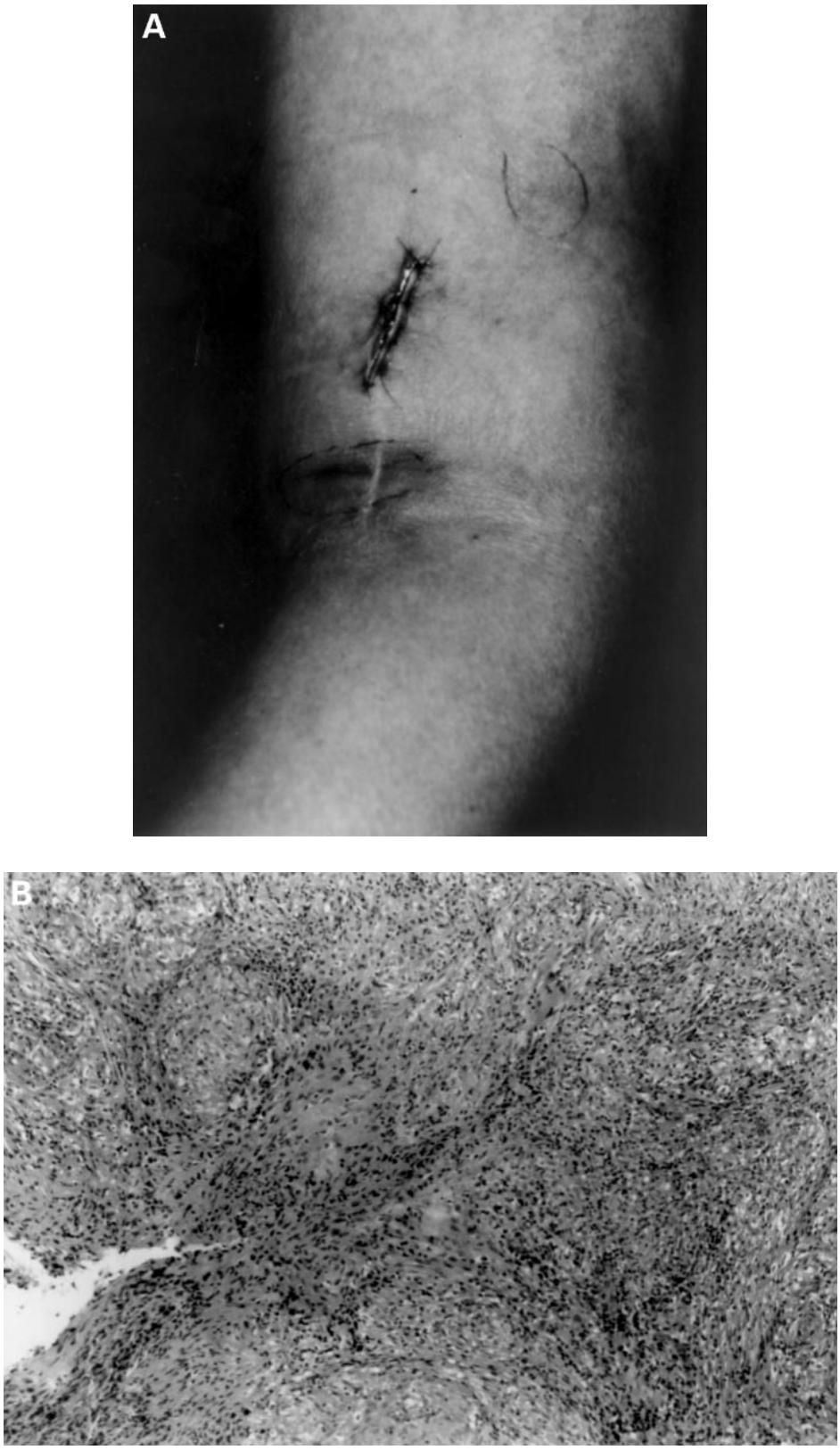

Fig 4 A 50-year-old widow with depression who had injected her forearm and antecubital fossa with vegetable matter. (A) appearance of the multiple subcutaneous masses. Note the biopsy site. (B) histological examination showing multiple granulomas (hematoxylin and eosin stain).

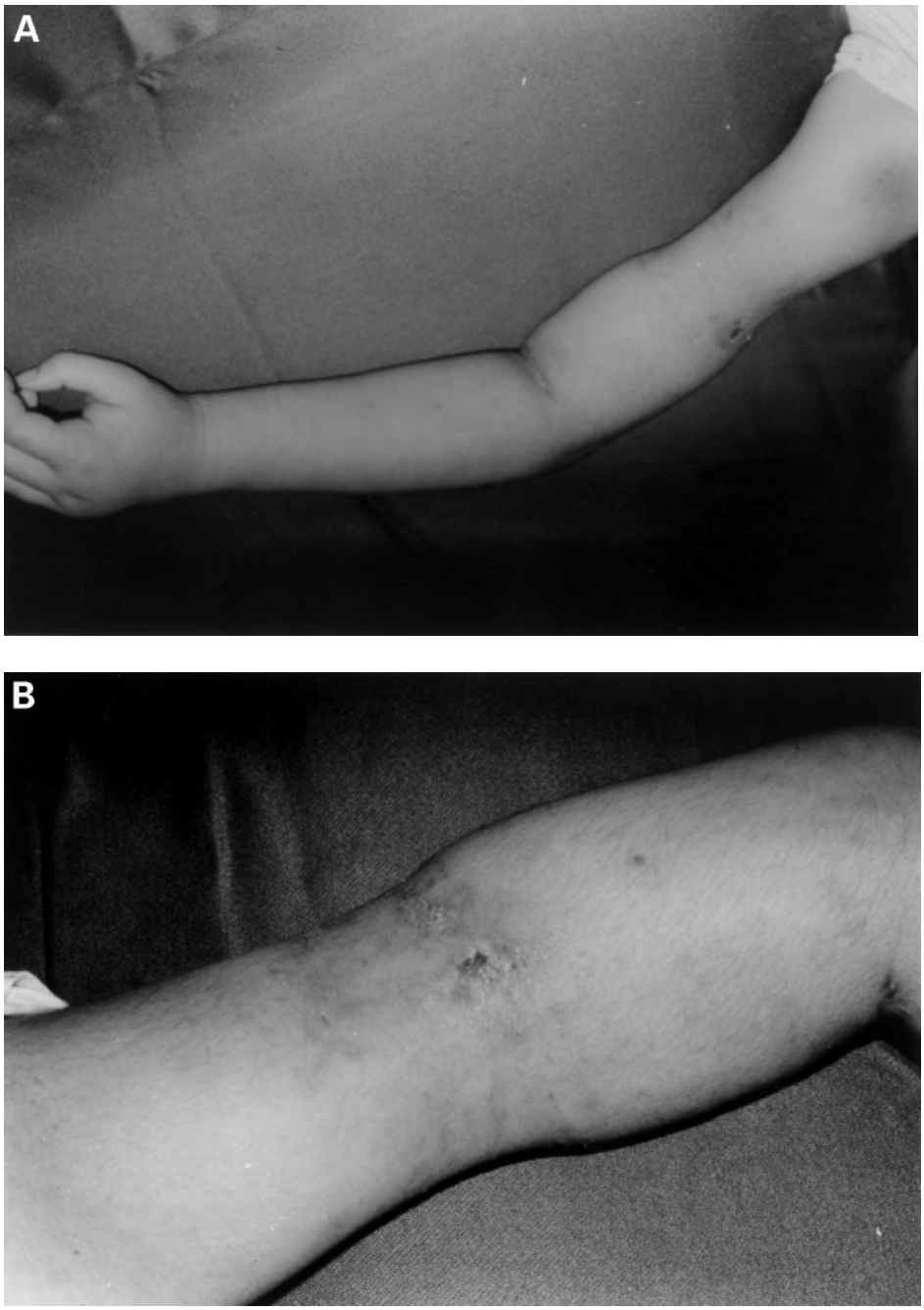

Fig 5 A 16-year-old girl referred to the hand clinic as a case of unilateral lymphoedema of the upper limb. (A) Clinical appearance of the limb. Note the demarcation line in the upper arm. (B) Close-up view of the demarcation line which was the site of tourniquet applied by the patient to induce oedema.

Hand posturing

This was the largest group and consisted of nine men and one woman (almost 99% of industrial and manual workers in Saudi Arabia are men). This group of ten patients was divided into two sub-groups. The group ($n = 3$) with long-standing psychological disease (depression or schizophrenia) presented with flexion contractures of the ulnar three fingers of one or both hands. There was no organic cause for these contractures and splinting only produced a temporary improvement. No surgery was performed and their treatment was always unsuccessful. The other group of patients consisted of seven manual or industrial male workers who had suffered "minor" hand injuries and had then presented with various hand postures (inability to extend fingers,

Fig 5 (C) Resolution of oedema 3 weeks after psychiatric consultation.

inability to flex fingers, stiff index finger, and thumb in palm deformity). These patients were malingerers feigning illness for secondary gain from compensation or lighter duties. The diagnosis in these patients was relatively easy to make because of the bizarre posture and because the patient resisted passive movements. Although the author did not directly confront these patients with the factitious nature of the hand posture, he indicated that the abnormal hand posture could not be explained by the previous minor hand injury and hence no compensation would be given. A short course of physiotherapy resulted in a "cure", with all patients returning to work.

DISCUSSION

Our study is the first investigation of factitious disorders of the upper limb in Saudi Arabia. The series was accumulated over 6 years from two large government hospitals and one large industrial injury hospital: hence the whole spectrum of factitious disorders was seen. Most previous papers have studied either a specific disorder (Frykman et al., 1983) or were based mainly on a population receiving worker's compensation benefits (Graham et al., 1999; Grunert et al., 1991).

In our study the predominance of women was marked, especially in the active mutilators, though hand posturing was mainly seen in men. Another interesting finding was the absence of Secretan's dorsal hand oedema. Furthermore, almost one-third of the women caused the injury by introducing a foreign body in the upper limb, a category that is rarely described in the hand surgery literature. The study also demonstrates that many cases do not fit into any of the recognized syndromes of factitious injury. For example, the foreign body introducers did not have the SHAFT personality, did not invent fanciful Münchhausen's stories and did not present with a wound. The classification offered in the current study (Table 2) includes three groups of active mutilators (wound inducers, foreign body introducers and oedema inducers). Passive mutilators are considered as a separate group and are defined as those who complain of factitious pain or numbness of the limb, and in whom there is no apparent secondary gain regarding employment or compensation. Finally, hand posturing is divided into two subgroups. The subgroup with major psychological illness usually presents with a clenched-fist and are very difficult to cure, as observed by Frykman et al. (1983). The other subgroup consists of employment or compensation-related malingerers who do not suffer with a major psychological illness and usually present with bizarre hand postures which are relatively easy to "cure". Grunert et al. (1991) noted that 80% of such patients returned to work, and a high success rate was also noted in the present series.

The diagnosis of factitious hand disorders in active and passive mutilators is often a lengthy process and is based on multiple observations by the physician, the psychologist and the hand therapist. Kasdan et al. (1998) identified several criteria that may help the hand surgeon in the diagnosis of these disorders. These include a history of multiple procedures, consultations with multiple physicians, a past history of psychiatric

treatment, being off work, crying with pain, a family history of disability and the absence of objective findings. However, some cases remain a diagnostic challenge (Friedman et al., 1988; Agris and Simmons, 1978). Once the diagnosis of a factitious disorder is made, the hand surgeon should give the patient the opportunity to "save face" and must not deprive him of the defences that maintain his emotional integrity. Furthermore, the patient should not be confronted with evidence of self-mutilation until a plan for social and psychiatric care is arranged. Untimely confrontation may result in more factitious illness, psychosis or a breakdown in the doctor–patient relationship (Friedman et al., 1988). The cost of care of these patients can be staggering (Barsky and Borus, 1995). Kasdan and Stutts (1995) estimated the total cost for one patient's care at $168,000. These high costs are not only because of time off work, but also because multiple surgical procedures are often performed. Some authors (Sennwald, 1998) believe that we should also consider the profile of the involved surgeons, since medical treatment needs a mutual consensus and there is no medical rationale for performing a large number of speculative procedures on a single patient. However, some of these patients are very intelligent and one of our patients had undergone six procedures under different surgeons. She then stopped injuring her hand for over 6 months, attended regular follow-up visits with her psychiatrist and hand surgeon, and therefore underwent release of her contracture. This was obviously the wrong decision, as the patient reverted to wound manipulation postoperatively.

Brown (1998) has recently pointed out that some hand surgeons only assess a hand or wrist that just happens to be connected to a patient, rather than seeing the person whose hand they are treating. The interface between upper extremity symptoms and psychiatric illness is much greater than hand surgeons appreciate and the time has come to educate ourselves regarding the signs and symptoms of psychiatric illness which present in the upper extremity (Stackhouse, 1998).

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