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CASE REPORT

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Munchausen syndrome with foreign-body granuloma mimicking rheumatic diseases

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Abstract A 37-year-old man was admitted to our hospital because of fever, polyarthralgia, and subcutaneous tumors. There was swelling of the bilateral wrists and ankles, and subcutaneous tumors over the bilateral elbow joints. Despite his complaints of multiple symptoms, clinical investigations failed to reveal any abnormality. Although laboratory parameters improved rapidly after steroid therapy, the symptoms remained unchanged, and there was an enormous discrepancy between the laboratory data and his symptoms. A biopsy specimen from one of the subcutaneous tumors revealed foreign-body granuloma associated with a foreign body fragment. Because the nursing staff later discovered that the patient had been carrying out self-injection, a diagnosis of Munchausen syndrome was made. Munchausen syndrome should be included in the differential diagnosis of rheumatic diseases.

Key words Factitious illnesses · Foreign body granuloma · Munchausen syndrome · Self-injection

Introduction

Munchausen syndrome is defined as a condition characterized by the feigning of symptoms of a disease or injury in order to undergo diagnostic tests, hospitalization, and medical treatment.[1] The name Munchausen comes from the German baron Karl Friedrich Hieronymous von Münchausen (1720–1797), who moved from city to city telling elaborate tales which had absolutely no basis in truth.[2] Since the first report of Munchausen syndrome, many case reports in various medical fields have documented the performance of unnecessary operations and the administration of dangerous drugs to affected patients.[3–6] However, rheumatologic manifestations in Munchausen syndrome are usually rare, and there seems to be little information in the rheumatological literature. We report the case of a patient who presented with symptoms mimicking a rheumatic disease such as Behçet's disease or Weber–Christian disease, but was finally diagnosed as having Munchausen syndrome with foreign-body granuloma caused by self-injection.

Case report

The patient was a 37-year-old man who had been hospitalized elsewhere from November 2001 because of lower abdominal pain. Although the pain had soon improved after the administration of antibiotics, he developed high-grade fever, polyarthralgia, and subcutaneous tumors in the middle of December 2001. Although several investigations failed to reveal the cause of these symptoms, a trial administration of prednisolone (PSL, 10 mg/day) was started, but the symptoms remained unaltered. The patient was moved to our hospital on March 27, 2002. His occupation was in construction, and his medical history revealed that he had undergone five operations over the previous 15 years (for a herniated intervertebral disk, a crush fracture of the right radius, right carpal tunnel syndrome, and two left thoracic sympathectomies). His family history was completely unknown because he had been cut off from his family since childhood.

On admission to our hospital, a physical examination revealed fever (body temperature 37.5°C), hen's egg-sized subcutaneous tumors with tenderness over the bilateral

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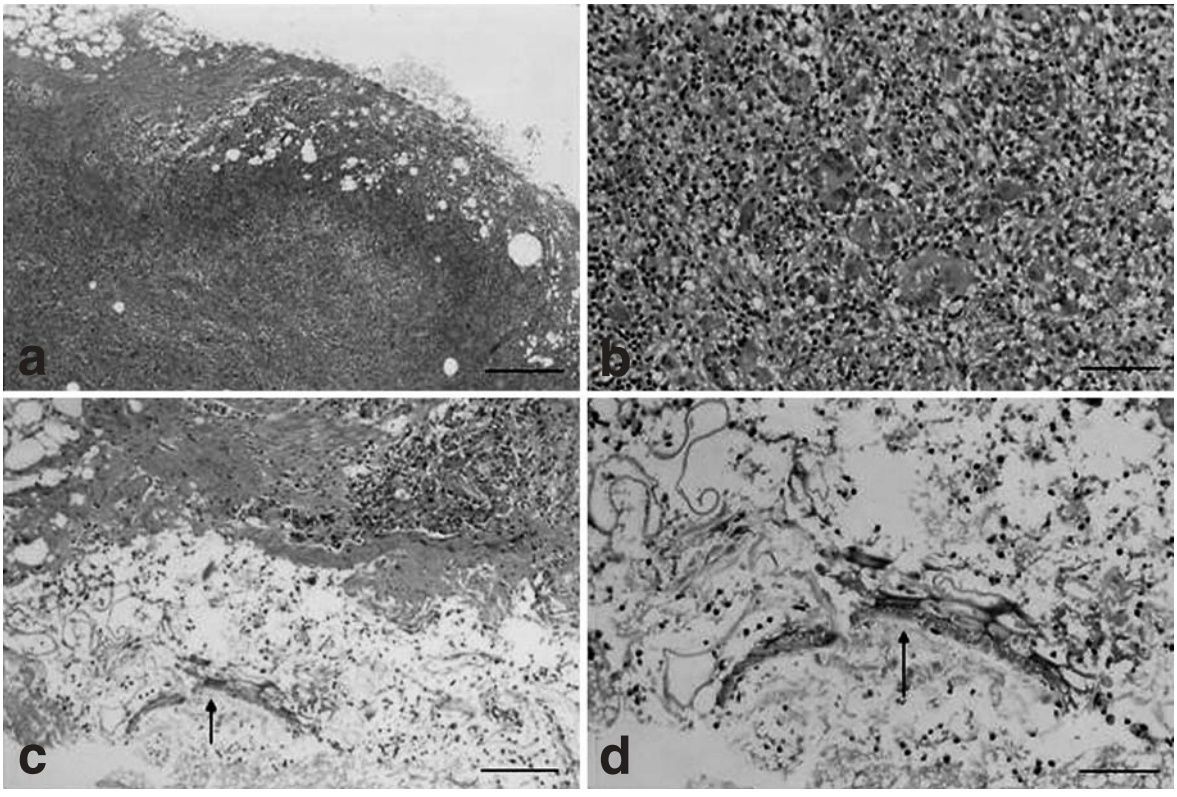

Fig. 1. a Histopathological appearance of the subcutaneous tumor, showing a foreign-body granuloma. **b** There are many giant cells and histiocytes and marked phagocytosis in the granuloma. **c** A foreign body (*arrow*) is evident at the site of panniculitis outside the granuloma. **d** The long, narrow foreign body (*arrow*) is divided into many compartments, including some eosinophilic material. H&E staining. *Bar* **a** 500 μm, **c** 250 μm, **b**,**d** 100 μm

elbow joints, and swelling of the bilateral wrists and ankles. Although he had tattoos on his back, shoulders, arms, and fingers, there was no skin eruption or tumor at these sites. Interestingly, the subcutaneous tumors were found to appear and disappear spontaneously after admission. Laboratory data, including leukocytosis (10200/μl), an elevated erythrocyte sedimentation rate (ESR, 78 mm/h), and a high C-reactive protein (CRP, 7.45 mg/dl) level, suggested systemic inflammation. Tests for antinuclear antibody, rheumatic factor, disease-specific autoantibodies, and antineutrophilic cytoplasmic antibodies all gave negative results. Complement components (C3, C4, and CH50) were within the normal ranges, and several hormonal tests revealed no abnormalities. Human leukocyte antigen (HLA) B51 was negative. Serum amyloid-A (SAA), protein (575 μg/ml), and several inflammatory cytokines such as interleukin (IL)-6 (366.03 pg/ml), interferon-γ (389 pg/ml), and IL-8 (19 pg/ml) showed significantly elevated levels. Since malignancy, infection, or rheumatic diseases were all clinical possibilities, the patient underwent a full investigation with X-ray roentgenography, computerized tomography, magnetic resonance imaging, echography, gastrointestinal fiberscopy, imaging of the small intestine, colonoscopy, and scintigraphy. However, none of these investigations revealed any abnormality. Cytological tests of sputum and urine revealed no malignancy, and various tumor markers were within normal limits. Cultures of urine, stools, and sputum grew no pathogenic bacteria, fungus, or mycobacterium. Thus, there was no apparent focus of infection, and the results suggested that malignancy and infection could be ruled out.

After admission, the patient began to complain of multiple symptoms such as polyarthralgia, headache, general fatigue, abdominal pain, back pain, constipation, nausea, appetite loss, and sleeplessness. We therefore performed a biopsy on one of the subcutaneous tumors, and the specimen obtained was found to be a foreign body granuloma with panniculitis (Fig. 1a). The granuloma contained many giant cells and histiocytes showing marked phagocytosis (Fig. 1b) resembling Behçet's disease or Weber–Christian disease. Interestingly, a foreign-body fragment was found at the site of panniculitis outside the granuloma (Fig. 1c). The foreign body was long and narrow, and divided into many compartments, including some eosinophilic material (Fig. 1d). Although the origin of the foreign body was investigated by pathologists, parasitologists, and some researchers, no conclusive result was reached. However, on the basis of its morphology, we speculated that the foreign body might be a piece of plant root.

Despite the lack of a definite diagnosis, we strongly suspected a rheumatic disease such as Behçet's disease, Weber–Christian disease, or unclassified connective tissue disease, although these were clinically atypical. Without waiting for the result of the biopsy, we started PSL admin-

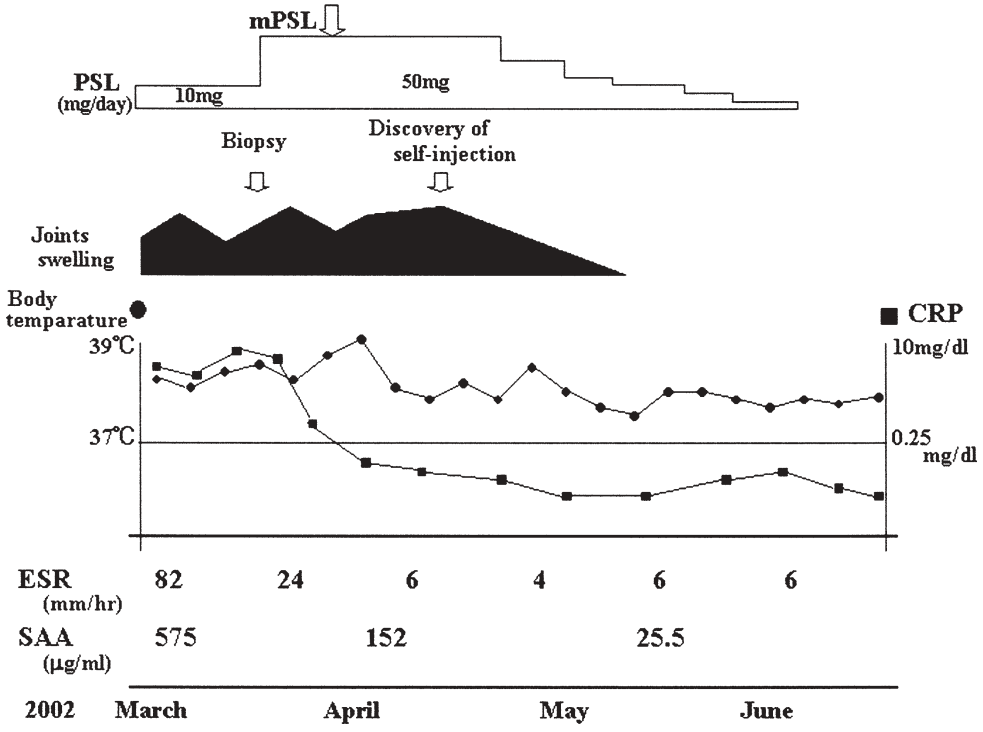

Fig. 2. Clinical course of the patient. There was enormous discrepancy between the laboratory data and his symptoms after treatment. *PSL*, prednisolone; *CRP*, C-reactive protein; *ESR*, erythrocyte sedimentation rate; *SAA*, serum amyloid-A

istration (50 mg/day) and methyl-PSL pulse therapy (1 g/day for 3 days) (Fig. 2) because of the severity of the symptoms. Although laboratory parameters such as CRP, ESR, and SAA protein improved rapidly, and the patient's body temperature decreased slightly after therapy, the symptoms were not altered, and in fact became exacerbated. Furthermore, the patient requested the insertion of a central vein catheter and intravenous hyperalimentation because of nausea and appetite loss. At this stage there was an enormous discrepancy between the laboratory data and the symptoms.

A diagnosis of Munchausen syndrome was considered, and the patient was confronted. However, he vehemently denied this. Subsequently, the nursing staff found by chance that the patient possessed an old syringe and needle containing cloudy liquid (Fig. 3a). He explained incoherently that the syringe was used to wash a wound on his foot. An analysis of the cloudy liquid was carried out by Triage (Sysmex, Kobe, Japan) with the patient's agreement, and the contents were found to be diazepam and amitriptyline hydrochloride (Fig. 3b). We concluded that the patient had taken these drugs, originally prescribed for his sleeplessness, and injected himself with them. The subcutaneous tumors appeared to be a local reaction to self-injection, while arthralgia and systemic inflammation were considered to be elicited by an adjuvant effect from the injected foreign body. This confirmed the diagnosis of Munchausen syndrome. A reduction of PSL was gradually achieved, since improvements in the laboratory data suggested that there was no further systemic inflammation. After the start of PSL reduction, the patient's symptoms showed no alteration, but no subcutaneous tumors or joint swelling were subsequently observed. He was transferred to another hospital on June 26, 2002, to receive psychiatric treatment.

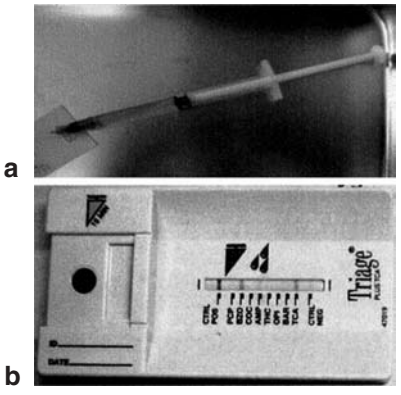

Fig. 3. **a** The old syringe and needle containing cloudy liquid found in the patient's possession. **b** The result of analysis revealed that the liquid contained diazepam and amitriptyline hydrochloride. *CTRL POS*, positive control; *PCP*, phencyclidine; *BZO*, benzodiazepines; *COC*, cocaine; *AMP*, amphetamines; *THC*, 11-nor-Δ^9-THC-9-carboxylic acid; *OPI*, opiates; *BAR*, barbiturates; *TCA*, tricyclic antidepressants; *CTRL NEG*, negative control

Discussion

In 1951, Richard Asher was the first to use the term Munchausen syndrome to refer to a peculiar subset of patients who repeatedly seek medical care for factitious illnesses.[2] The main feature of this syndrome is the simulation

or fabrication of physical symptoms and signs, psychiatric symptoms and signs, or both, with no apparent motivation other than to assume the role of a patient.[1] The patient presents a characteristic triad: apparently acute but factitious disorders, migration from hospital to hospital resulting in unnecessary tests and treatments, and a fabricated medical history. A review of the patient's family and personal background might reveal a history of childhood separation and emotional neglect or abuse. In this context, their actions could be viewed as an abnormal means of eliciting care. Recent life-events might also be important, the factitious behavior allowing the patients to escape the reality of their current situation. The past and current family and personal background of our patient undoubtedly played a role in the occurrence of this syndrome.

The diagnosis of Munchausen syndrome is remarkably difficult, and is often not considered during the initial assessment. The diagnosis can be made by excluding various possibilities after numerous tests have produced normal results. If a rheumatic disease is suspected because manifestations similar to rheumatic diseases are present, it is especially difficult to reach the proper diagnosis at the initial assessment because rheumatologic manifestations in this syndrome are rare, and rheumatic diseases are systemic, producing various symptoms and involving multiple organs. Therefore, Munchausen syndrome should be considered in the differential diagnosis when some symptoms similar to rheumatic diseases are present but there is no evidence of autoimmunity. In our case, since the patient's rheumatic disease-like symptoms were extremely severe, and laboratory data clearly revealed a systemic inflammation, we carried out steroid therapy before reaching a definite diagnosis. Nevertheless, we had considered the possibility of Munchausen syndrome. Neglecting the possibility of Munchausen syndrome could delay a proper diagnosis and lead to expensive tests, as well as meaningless and dangerous treatments. However, when considering Munchausen syndrome, it should be remembered that a genuine physical illness underlying the syndrome might also be presented.

Munchausen syndrome is frequently refractory to treatment, and its management has received little attention in published work. In fact, there has been no study of its treatment. Psychiatric treatment is the mainstay, but rarely has a favorable outcome, and may itself cause problems because a stay in a mental health unit might provide the patient with fertile new areas of symptomatology to exploit. Finally, the patient may elude meaningful therapy by abruptly leaving hospital, or failing to keep follow-up appointments. Our case was presumed to be refractory to treatment because of the lack of sick consciousness and the length of the illness. The most important aspect of management for this syndrome is early recognition by physicians that the illness is factitious.

In conclusion, Munchausen syndrome is a rare, but often severe, destructive disorder that can be diagnosed by exclusion. The first step toward a proper diagnosis is to ensure that no genuine physical illness is present. Munchausen syndrome should be included in the differential diagnosis of rheumatic diseases.

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