Subcutaneous emphysema (SE) is a condition that shows aberrant gas in the soft tissue. It can be life-threatening when associated with aerodigestive tract perforation or gas gangrene, but has a benign course when caused by recent skin trauma, including pneumatic tool injury [1], skin biopsy [2] and cryotherapy [3]. Self-inflicted or factitious SE (FSE) is very rare. About a dozen cases are documented in the English-language literature. Patients with FSE often seek medical attention from different medical specialties according to specific somatic complaints, thus resulting in a waste of medical sources and delay of psychiatric referral. The factitious nature of SE is usually suspected only after exclusion of an underlying medical emergency. We report a complex case of recurrent SE in a young woman with underlying psychiatric disorder.

**CASE PRESENTATION**

We were consulted in June 2008 by an anesthesiologist to evaluate a 29-year-old woman who presented with recurrent painful soft tissue swelling of the anterior chest wall, abdomen and neck over several months. Factitious disease was suspected because of the negative medical history of identifiable visceral perforations or gas-forming infections and the presence of multiple bizarre linear scars on the arm. This was supported by finding multiple recent puncture marks on the neck, chest and upper extremities on close inspection. The patient’s history was remarkable for multiple stressful events in her life, frequent job changes, substance abuse and suicide attempts. Psychiatric evaluation revealed adjustment disorder with depression and anxiety, cluster B personality with bipolar II disorder and substance abuse. During the 1-year period, she had 20 visits to our ER for attacks of SE affecting various parts of her body. This case illustrates that self-infliction should be suspected in a patient presenting with medically unexplained recurrent SE and a hollow history, and one should search for puncture marks to support the diagnosis.
chest wall, abdomen and neck over the past several months. She was admitted to our General Surgery via the Emergency Room (ER) in May 2008 for treatment of SE. Her past history was remarkable for similar recurrent painful attacks (first episode at 4 years of age, according to the patient) and had multiple surgical treatments at other hospitals. Most of the previous attacks were not associated with constitutional symptoms. She had undergone laparoscopic surgery for uterine adenomyosis at a nearby hospital in April 2008. One week postoperatively, she returned to that hospital with abdominal pain; a computed tomography (CT) scan revealed air collection in the abdominal wall. Her symptoms were relieved by insertion of Penrose drains in the periumbilical area. An iatrogenic cause of SE was suspected. She was transferred to our ER for further management.

During hospitalization, the patient complained of new pain with subcutaneous crepitus extending to her right anterior chest. A CT scan revealed massive subcutaneous air over the neck, chest and abdomen without pneumothorax, aerodigestive tract perforation (Figures 1A and 1B). There was no evidence of gas-forming infection. Another Penrose drain was placed on the chest wall (Figure 1C). In a psychiatric consultation, she reported having a stressful and insecure life in the past 6 years living with a boyfriend who was a drug abuser with violent behavior and multiple criminal records, and who had threatened to take away her daughter. She could not cope well at work and had frequent job changes (27 jobs in 1 year). Under the tentative diagnosis of adjustment disorder with depressive and anxious mood and insomnia, antidepressant (paroxetine) and benzodiazepine (alprazolam and lorazepam) were prescribed. However, a second consultation was requested a week later for episodes of emotional instability. Cluster B personality with bipolar II disorder was added to the diagnosis, and valproic acid was started. After discharge from our hospital, she returned to our ER several times for recurrent painful attacks, and was referred to the Pain Clinic.

During the dermatology consultation, she experienced several bouts of pain with agitated mood and outbursts of crying. On examination, crepitus could be

Figure 1. Computer tomography scan revealed the presence of massive subcutaneous air over (A) the chest (arrow) and (B) the abdomen (arrow) without evidence of pneumothorax, aerodigestive tract perforation. (C) Penrose drains are placed on the neck and right anterior chest wall for extensive subcutaneous emphysema.
felt in the neck, anterior chest and arms. Multiple linear scars from previous operations were found on various parts of her body and several old, bizarre linear scars on the antecubital area (Figure 2A). Close inspection revealed several tiny red spots and atrophic scars resembling recent and old needle puncture marks near or at the areas affected by emphysema (Figures 2B and 2C). When asked, she attributed these marks to venous punctures by the hospital staff. A factitious cause was thus suspected.

In a psychiatric evaluation in November 2008, she admitted substance abuse of amphetamine and sleeping pills since 2001, suicide attempts by wrist cutting at 18 years of age, and heroin injection starting in October 2008. Methadone treatment was initiated but was maintained for only 3 months. In the following 6 months, she was often unemployed and in financial difficulty. She continued to complain of insomnia, stress, weight loss, and recurrent painful attacks at different sites for which she made frequent visits to our ER and Pain and Psychiatry Clinics for symptom relief. Various drugs including lorazepam were prescribed at different times mainly for anxiety and insomnia, and then she was switched to antipsychotics (olanzapine) along with flunitrazepam (January 2009), valproic acid, trazodone, and lorazepam (March 2009) and quetiapine, chlor Diazepoxide, trazodone, and lorazepam (May 2009). For pain control, she was given ketorolac or tramadol injection mostly when she visited the ER. Morphine 40 mg and tramadol 200 mg per day were prescribed regularly from February 2009 onwards.

Despite these medications, she had 12 more visits to the ER, two admissions to Surgery, and about a dozen visits to the Pain and Psychiatric Clinics in 3 months (February to May, 2009) for recurrent painful attacks of SE affecting various parts of her body. In February, she had pain in the neck, left arm and palm accompanied by shortness of breath and fever. Radiographic examination revealed air in the forearm, wrist, hand and mediastinum. Some needle puncture marks were noticed on the left hand and elbow during a psychiatric consultation, but the patient explained that they were scratch marks. She had several visits to ER in the following month for separate episodes of SE in the right foot, right leg, right neck, and right mandibular area, confirmed by CT scan. This time, she denied a prior history of SE. In addition to SE attacks, she had one ER visit for a suicidal attempt (taking 40 sleeping pills at one time), and another for cutting wounds in the arms and back that she claimed to be caused by her husband. Repeated assistance by social workers were met with resistance and proved to be useless.

**DISCUSSION**

We have described a case of chronic, recurrent SE in a young woman with many stressful life events and an underlying adjustment disorder, cluster B personality with substance abuse and suicide attempts. The painful swelling attacks were migratory as she produced SE.
in various parts of her body over a period of 1 year. Iatrogenic SE had been considered by general surgeons at first due to her recent history of abdominal surgery and the involvement of the periumbilical area. However, a suspicion of self-infliction was soon raised during the dermatology consultation because many recent and old needle puncture-like marks were distributed over the crepitus areas that were within her reach. The presence of bizarre linear scars on the arms also attested to her past suicide attempts and self-mutilation. During the 1-year period, the patient had three hospital admissions, 20 ER visits, and about 30 visits to the Pain and Psychiatric Clinics where she sought help from various physicians, mainly for treatment to relieve pain and insomnia. The patient seemed evasive and resistant to intervention by social workers. We suspect that, because of her substance abuse, she might have been feigning more frequent pain attacks in order to obtain more opioid analgesics from our hospital, and possibly from other hospitals.

Although factitious disorder (FD) is a psychiatric condition, it challenges various medical specialties [4]. FD is categorized as an Axis I DSM-IV condition but has been reported to be associated with depression, schizophrenia, and personality disorders. FD is diagnosed when there is intentional production or feigning of physical or psychological signs or symptoms where the incentive is to assume the role of a sick person, and external incentives for the behavior are absent [5]. The main differential diagnosis is malingering, in which external incentives, such as monetary reward, relief from occupational or other social responsibility, or avoiding standing trial, are present for the intentional behavior. Munchausen syndrome is a special type of FD in which patients deliberately produce signs and symptoms of a disease supported by elaborately contrived but just plausible medical histories and allow themselves to be investigated extensively and treated for no apparent reason other than to baffle the physician [6].

Among FDs, factitious dermatitis or dermatitis artefacta (DA) is well known to dermatologists because of the skin lesions produced. DA can manifest in various types of skin lesions and affect various parts of the body. Clinically, the lesions may mimic other conditions, such as infected ulcers, cellulitis and pyoderma gangrenosum. Nielsen et al recently reported a series of 57 cases of DA (mean age = 39 years; range, 12–86 years; female-to-male ratio = 2.8:1) diagnosed in a Denmark university hospital over 20 years (1982–2002) [7]. In that series, about 75% of the patients were jobless or on sick leave, 18% had alcohol or substance abuse, and 18% had a psychiatric diagnosis. The most common “objective” skin lesions were ulcers or erosions, excoriations and localized erythema, and 88% of the patients had multiple lesions.

DA often remains undiagnosed until the bizarre appearance of the skin lesions has been recognized along with nonspecific histopathological findings and unrevealing systemic work-ups. Clues to the factitious nature of the lesions include: occurrence in areas readily accessible to the patient, geometric patterns or angulated borders of the lesions surrounded by completely healthy skin, and bizarre morphology not conforming to typical presentations of known dermatoses. Patients with DA produce lesions intentionally and have control in planning the full course of the factitious illness [8]. Patients typically attribute the lesions to other reasons and deny self-infliction. In the series by Nielsen et al, in which 30 patients were confronted with the possibility of self-infliction, two-thirds denied this, and only one agreed to see a psychiatrist. Most patients have a personality disorder with borderline features and are vulnerable to life events [8,9]. Many of these features were present in our patient. DA is unlike other self-inflicted skin diseases such as trichotillomania and neurotic excoriations. The latter are associated with obsessive-compulsive disorder characterized by preoccupied thoughts and uncontrollable impulse or behaviors, while DA is not.

FSE appears to be very rare. We found 12 cases reported in the English-language literature [8,10–20]. Including our patient, there were three males and 10 females with ages ranging from 2 months to 44 years (mean 23.4 years) (Table). Most patients were adolescents or young adults. The patients typically presented with unexplained regional subcutaneous crepitus without serious constitutional medical problems. Most had an underlying psychiatric condition or other relevant personal history, including personality disorders (5 cases), depression, self-mutilation behavior, suicide attempts, or problems with marriage or interpersonal relationships. The body sites affected included the upper extremities (7 cases), periorbital areas and face (5 cases), neck and chest (2 cases), abdomen (1 case) and lower extremity (3 cases). In the most severe case, the self-infliction resulted in proptosis, blindness, and eventual exenteration of the right eye [13]; the final
Recurrent factitious subcutaneous emphysema

Diagnosis of Munchausen’s syndrome in this patient was made only after extensive investigation to evaluate the pneumo-orbit of her remaining left eye 10 years later. The youngest patient was a 2-month-old infant. The infant presented with SE involving the neck and chest as a result of child abuse [16]. Child abuse or Munchausen’s syndrome by proxy needs to be considered in cases of young children.

It should be noted that the 12 cases of FSE had been reported by various medical specialties, including otolaryngology, orthopedics, pediatrics, emergency medicine, surgery, internal medicine and dermatology (Table). This may be because FSE manifests no obvious skin lesions and most patients present themselves to the emergency services or non-dermatology specialists with symptoms that cause a concern as a medical emergency. In contrast, patients with DA often present to dermatologists directly because there are obvious skin lesions. Only one of the 12 FSE cases was reported by dermatologists and it was mild clinically [8]. We would not have the opportunity to see the present patient if the anesthesiologist, who managing her pain, did not ask one of us (Dr J.Y.Y. Lee) to see her.

Despite the rarity of FSE, we did see another case in our clinic 2 years ago. The patient was a 16-year-old girl, who visited our clinic for recurrent painful attacks over the chest and forearms and was initially diagnosed as having herpes zoster. Crepitus in both forearms was found this time, and the diagnosis of SE was confirmed by radiography. Multiple needle puncture marks were discernible on the forearms. In addition, bizarre linear scars were also noted over bilateral upper arms.

Table. Summary of 13 patients with factitious subcutaneous emphysema reported in the English-language literature

<table>
<thead>
<tr>
<th>Case</th>
<th>Age (yr)</th>
<th>Sex</th>
<th>Sites</th>
<th>Psychiatric disorder or other relevant personal history</th>
<th>Reporting medical specialty, year [Reference]</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>28</td>
<td>F</td>
<td>Arm</td>
<td>Hysterical personality, unhappy marriage and multiple miscarriages</td>
<td>Medicine, 1971 [10]</td>
</tr>
<tr>
<td>2</td>
<td>15</td>
<td>F</td>
<td>Arm</td>
<td>Hysterical conversion reaction, difficulty coping with family and peers</td>
<td>Pediatrics, 1976 [11]</td>
</tr>
<tr>
<td>3</td>
<td>20</td>
<td>M</td>
<td>Arm</td>
<td>Jobless, self-mutilation behavior; syringe found</td>
<td>Surgery, 1977 [12]</td>
</tr>
<tr>
<td>4</td>
<td>23</td>
<td>F</td>
<td>Face, periorbital, foot</td>
<td>Depression; caught hiding air-filled syringes</td>
<td>Otolaryngology, 1983 [13]</td>
</tr>
<tr>
<td>5</td>
<td>15</td>
<td>M</td>
<td>Face, periorbital</td>
<td>Divorced family</td>
<td>Pediatrics, 1984 [14]</td>
</tr>
<tr>
<td>6</td>
<td>40</td>
<td>F</td>
<td>Hand</td>
<td>None mentioned</td>
<td>Orthopedics, 1994 [15]</td>
</tr>
<tr>
<td>7</td>
<td>0.16</td>
<td>F</td>
<td>Neck, chest</td>
<td>Child abuse</td>
<td>Emergency medicine, 1997 [16]</td>
</tr>
<tr>
<td>8</td>
<td>36</td>
<td>F</td>
<td>Face, periorbital</td>
<td>Angry and lost to follow-up when confronted with question of self-infliction by a psychiatrist</td>
<td>Otolaryngology, 1998 [17]</td>
</tr>
<tr>
<td>9</td>
<td>13</td>
<td>F</td>
<td>Arm</td>
<td>Problems at school and home</td>
<td>Dermatology, 2006 [8]</td>
</tr>
<tr>
<td>10</td>
<td>20</td>
<td>F</td>
<td>Face, periorbital</td>
<td>Caught injecting air into the face during hospitalization; transferred to psychiatric care</td>
<td>Otolaryngology, 2007 [18]</td>
</tr>
<tr>
<td>11</td>
<td>21</td>
<td>M</td>
<td>Thigh</td>
<td>Early childhood emotional and physical abuse; borderline personality; “factitious disorder” diagnosed.</td>
<td>Orthopedics, 2008 [19]</td>
</tr>
<tr>
<td>12</td>
<td>44</td>
<td>F</td>
<td>Arm</td>
<td>Borderline personality disorder</td>
<td>Orthopedics, 2008 [20]</td>
</tr>
<tr>
<td>13</td>
<td>29</td>
<td>F</td>
<td>Chest, abdomen, neck, arms, foot, leg, face</td>
<td>Adjustment disorder, cluster B personality disorder, bipolar II, substance abuse, suicide attempts</td>
<td>Dermatology, present case</td>
</tr>
</tbody>
</table>
The clinical course of FDs is basically determined by the underlying psychopathology [8]. Although psychiatric consultation and management is mandatory for most patients with FD, it is often difficult to introduce psychological issues to patients and keep them on regular psychiatric treatment. Frank confrontation may precipitate psychosis or even suicide. Nevertheless, failure to confront the patient may reinforce such behavior and allow continuing exploitation [10], as is well exemplified by the present case. In a systemic review of 32 case reports and 13 case series of FD, Eastwood and Bisson found no significant differences in the outcome between confrontational and non-confrontational approaches, nor between with and without psychotherapy or psychiatric medication [4]. The management of our patient proved very challenging and treatment was ineffective. In our opinion, an integrated intervention involving appropriate medical specialties, nurses and social workers, social welfare assistance, family support and an enforced drug detoxification program would have been required to provide more effective help to the patient.

In conclusion, we report a very complex case of recurrent FSE in a young woman with an underlying adjustment disorder, cluster B personality, bipolar II disorder and substance abuse. When a patient presents with an unexplained recurrent SE with a hollow history, self-infliction should be suspected, and close inspection for identifiable puncture marks should be undertaken.

ACKNOWLEDGMENT

The authors are grateful to Mr Roderick Fletcher for checking the English in this manuscript.

REFERENCES

反覆人為性皮下氣腫：
一個錯綜複雜的年輕女性病例報告及文獻回顧

李宜珮 李玉雲
國立成功大學 醫學院及附設醫院 皮膚部

皮下氣腫可導因於先前的外傷、手術、內臟器官破裂或產氣菌造成的嚴重感染。人為性皮下氣腫非常少見，截至目前為止，共有 12 個病例不同專科領域的英文期刊被報導過。大多數的病人有精神疾病或相關病史。人為性皮下氣腫的診斷及治療相當困難。我們要報告一個臨床病程複雜的人為性皮下氣腫的新病例。該病例為 29 歲年輕女性，在過去的數個月於胸部、腹部及頸部出現反覆疼痛的軟組織腫脹。由於病史中並無內臟器官破裂或產氣菌感染等的證據，而且在兩側手臂上出現數個奇怪直線型的疤痕，所以才懷疑是否有人為導因，接著近看才發現於頸部、胸部及上肢出現數個針扎的痕跡。病人的過去病史包括承受數次生活的壓力事件、工作不穩定、藥物濫用及自殺意圖。精神科的評估診斷包括有適應性情緒疾患、憂鬱焦躁傾向、邊緣性人格異常、躁鬱症及物質濫用。在 1 年之中，她因為全身各部位反覆的皮下氣腫而急診求診 20 次。這個病例顯示出，當一個病人出現無法解釋的反覆性皮下氣腫時，再加上空洞的病史時，就應該尋找是否有針扎傷痕來支持這項診斷。

關鍵詞：人為皮膚疾病，人為造成的異常，反覆，皮下氣腫
（高雄醫誌 2010;26:377–83）

收文日期：98 年 7 月 22 日
接受刊載：99 年 1 月 4 日
通訊作者：李玉雲醫師
國立成功大學醫學院及附設醫院皮膚部
台南市勝利路 138 號