Case Report

Sweet's syndrome: a very rare association with pulmonary tuberculosis

Partha Sarathi Karmakar, Pasang Lahmu Sherpa, Achintya Narayan Ray, Bikram Kr. Saha, Tuhin Santra, Sanchita Saha, Indranil Chakrabarti

Department of Medicine, North Bengal Medical College and Hospital, Siliguri, West Bengal, India

Abstract

Mycobacterium tuberculosis infection is a common infection in developing countries, including India. It can induce several cutaneous reactions such as erythema nodosum, and erythema induratum; however, association of tuberculosis with Sweet's syndrome (also known as acute febrile neutrophilic dermatosis) is extremely rare. Here we present an interesting case of sputum-positive pulmonary tuberculosis with Sweet's syndrome. A 55-year-old female who was receiving a regimen of four antitubercular drugs (isoniazid, rifampicin, pyrazinamide, ethambutol- HRZE) for six weeks for sputum-positive pulmonary tuberculosis developed new onset high-grade fever for 15 days along with multiple reddish brown plaques and nodules involving the face as well as all four limbs of the body. Histopathology of the skin lesion was suggestive of Sweet's syndrome. The patient responded well to immunosuppressive steroid therapy.

Key words: acute febrile neutrophilic dermatosis; erythema nodosum; pyoderma gangrenosum

J Infect Dev Ctries 2013; 7(5):417-420.doi:10.3855/jidc.2606

(Received: 22 February 2012 – Accepted: 28 Septemer 2012)

Introduction

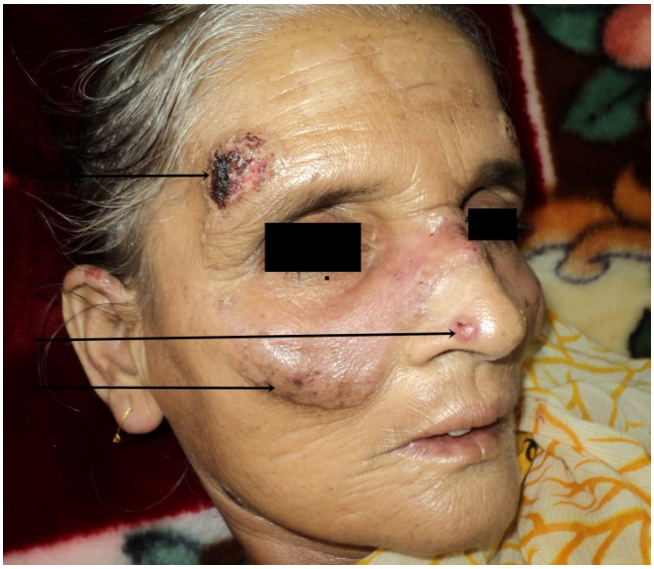
Sweet's syndrome (SS) was named after Dr. Robert Douglas Sweet from Plymouth, England, who first described this condition in 1964. It was also known as Gomm-Button disease in honour of the first two patients of Sweet's syndrome diagnosed by Dr. Sweet [1]. Sweet's syndrome can be classified based upon the clinical setting in which it occurs: classical or idiopathic Sweet's syndrome, malignancy associated Sweet's syndrome, and drug-induced Sweet's syndrome [2]. Sweet's syndrome is a reactive phenomenon and it should be considered a skin manifestation of systemic disease. Nearly 20% of all cases are malignancy related, predominantly haematological (acute myelogenous leukemia) [2]. The drug-induced variant of Sweet's syndrome is most frequently observed following administration of granulocyte-colony stimulating factor (G-CSF) [3,4]. Other commonly used drugs reported to cause Sweet's syndrome are antibiotics such as trimethoprim-sulfamethoxazole [5], nitrofurantoin [6], antiepileptic drugs such as carbamazepine [7], diazepam [8], diuretics such as furosemide [9], non-steroidal anti-inflammatory drugs such as diclofenac [10], and celecoxib [11]. Sweet's syndrome has also been reported following infections of the upper respiratory tract [12,13] and the gastrointestinal tract [14]. *Mycobacterium* cervical lymphadenitis associated with Sweet's syndrome has been documented in only a few reports [15,16], but sputum-positive pulmonary tuberculosis showing Sweet's syndrome is a very uncommon association. A case of Sweet's syndrome with pulmonary tuberculosis and cervical cancer has also been reported [17].

Apart from Sweet's syndrome, pyoderma gangrenosum and subcorneal pustular dermatosis are also considered to be neutrophilic dermatosis, as all these skin lesions have a dense inflammatory infiltrate consisting of mature polymorphonuclear cells [18].

Case report

A 55-year-old non-diabetic, non-hypertensive female patient was admitted in our institute with complaints of a high-grade continuous fever lasting 15 days along with the development of multiple reddish-brown elevated skin lesions mainly involving the face and all four limbs (Figures 1 and 2). The patient also experienced respiratory distress and chest pain for the same duration. She was receiving a regimen of four anti-tubercular drugs (isoniazid, rifampicin, pyrazinamide, ethambutol – HRZE) for the last six weeks for sputum-positive pulmonary tuberculosis.

Figure 1. Multiple lesions of Sweet's syndrome on face



Several types of lesions of Sweet's syndrome on face showing a red-brown juicy plaque (right cheek), a nodule (near tip of nose), and an ulcerated lesion (above right eyebrow)

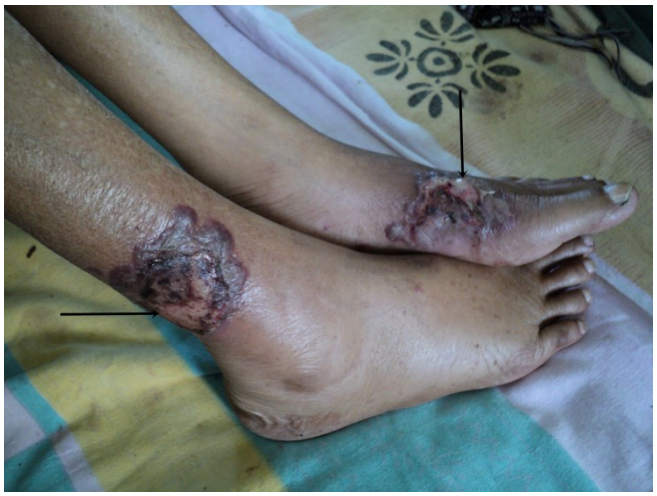
On examination, the patient was found to have poor nutritional status, a pulse rate of 98 beats per minute, blood pressure of 100/60 mm of Hg, a respiratory rate of 26 breaths per minute and a moderately raised temperature. There were multiple red-brown juicy plaques and nodules involving the face and the extensor surface of the upper and lower extremities, most of which became ulcerated over the next two to three days of hospital stay. Systemic examination revealed scattered coarse crepitations on both halves of the chest. There was no lymphadenopathy and no hepatosplenomegaly.

Investigations revealed hemoglobin 9.9 g/dl, total leukocyte count 25,400/mm3 with 90% polymorphonuclear cells (PMNs), platelet count 354,000/mm3, and erythrocyte sedimentation rate (ESR) 80 mm in the first hour.

Biochemical examinations including blood sugar, renal profile and liver function test were within normal limits. Routine urine and microscopic examinations were within normal limits. Chest X-ray showed bilateral patchy opacities suggestive of pulmonary tuberculosis (Figure 3).

Dermatological opinion was taken and prednisolone 40 mg/day along with amoxicillin clavulanate by injection was instituted on the basis of suspicion of Sweet's syndrome after taking a biopsy from a skin lesion of the right hand.

Figure 2. Lesions of Sweet's syndrome in legs



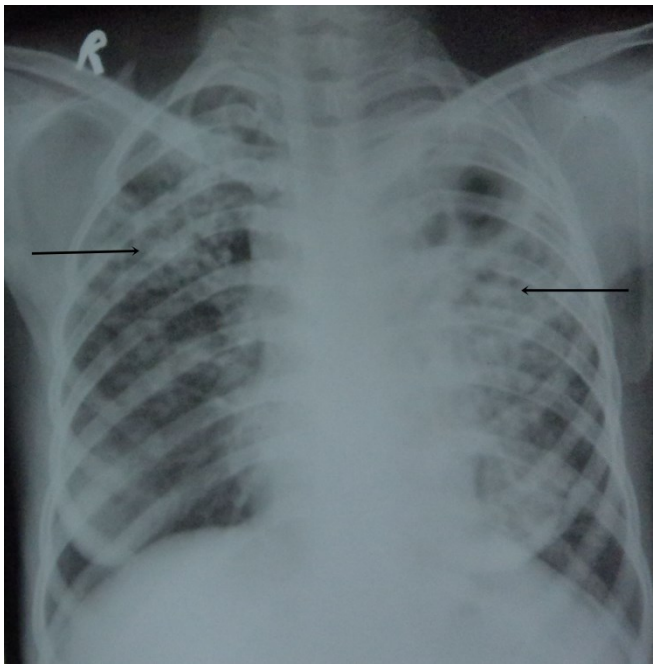
Ulcerated and crusted lesions of Sweet's syndrome in both legs

Significant improvement in the form of regression of skin lesions along with the patient's general status were noticed within three to four days of therapy. Unfortunately, five days later the patient suddenly had profuse hemoptysis and died of asphyxiation. Histopathological features from skin lesions showed epidermal and subepidermal (reticular dermis) dense infiltration of acute inflammatory cells (PMNs) suggestive of Sweet's syndrome (Figure 4).

Discussion

Sweet's syndrome is predominantly a female disease. In a study of 16 cases of Sweet's syndrome, 82% of the patients were female, and although previous infection was reported as contributing as a causal factor for Sweet's syndrome in 31% of all cases, and only one of these patients had primary pulmonary tuberculosis [10]. An extensive internet search for an association between tuberculosis and Sweet's syndrome showed only a few case reports, most of which were extra-pulmonary. To the best of our knowledge, this is the first case of an association of pulmonary tuberculosis and Sweet's syndrome reported from a developing country such as India. The possibility of antituberculosis drug (ATD)-induced Sweet's syndrome in this case cannot be ruled out; however, its cure in some anecdotal case reports with conventional anti-inflammatory and/or immunosuppressive therapy without interruption of ATD strongly goes against this hypothesis of ATD-induced Sweet's syndrome [16,17]. There are only a few case reports showing an association between Sweet's syndrome and tuberculosis both before and

Figure 3. Chest X-ray PA view



Bilateral patchy fluffy shadows (left > right) suggestive of pulmonary TB

after initiation of ATD therapy for a variable period of time ranging from two to nine months [15,17,19].

The most effective treatment of Sweet's syndrome is immunosuppressive therapy, most frequently with a systemic steroid. Other modalities of therapy include colchicine, indomethacin, clofazimine, cyclosporine, and dapsone. Recently there have been reports of patients with Crohn's disease and Sweet's syndrome responding to immunosuppressive therapy with infliximab [20, 21].

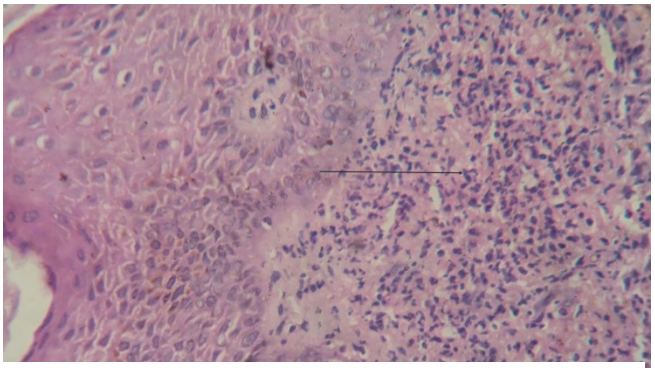
Conclusion

This is a well-documented clinical and histological case report of a recognised association of Sweet's syndrome in the course of pulmonary tuberculosis. This skin lesion should be included as one of the associated reactionary skin lesions of common diseases such as tuberculosis.

References

1. Sweet RD (1964) An acute febrile neutrophilic dermatosis. Br J Dermatol 76: 349-356.
2. Cohen PR (2007) Sweet's syndrome-a comprehensive review of an acute febrile neutrophilic dermatosis. Orphanet J Rare Dis 2: 34.
3. Shimizu T, Yoshida I, Eguchi H, Takahashi K, Inada H, Ando A, Kato H (1996) Sweet syndrome in a child with aplastic anemia receiving recombinant granulocyte colony-stimulating factor. J Pediatr Hematol Oncol 18: 282-284.

Figure 4. Histopathology of skin lesion



(Magnification x 400) Histopathology of skin lesion; arrow shows dense dermal infiltration of neutrophils (PMNs)

4. Garty BZ, Levy I, Nitzan M, Barak Y (1996) Sweet syndrome associated with G-CSF treatment in a child with glycogen storage disease type Ib. Pediatrics 97: 401-403.
5. Walker DC and Cohen PR (1996) Trimethoprim-sulfamethoxazole-associated acute febrile neutrophilic dermatosis: case report and review of drug-induced Sweet's syndrome. J Am Acad Dermatol 34: 918-923.
6. Retief CR and Malkinson FD (1999) Nitrofurantoin-associated Sweet's syndrome. Cutis 63: 177-179.
7. Sitjas D, Cuatrecasas M, De Moragas JM (1993) Acute febrile neutrophilic dermatosis (Sweet's syndrome). Int J Dermatol 32: 261-268.
8. Guimera FJ, Garcia-Bustinduy M, Noda A, Saez M, Dorta S, Sanchez R, Martin-Herrera A, Garcia-Montelongo R (2000) Diazepam-associated Sweet's syndrome. Int J Dermatol 39: 795-798.
9. Govindarajan G, Bashir Z, Kuppuswamy S, Brooks C (2005) Sweet syndrome associated with furosemide. South Med J 98: 570-572.
10. Ginarte M, Garcia Doval I, Toribio J (1997) Sweet's syndrome: a study of 16 cases. Med Clin (Barc) 109: 588-591.
11. Fye KH, Crowley E, Berger TG, LeBoit PE, Connolly MK (2001) Celecoxib induced Sweet's syndrome. J Am Acad Dermatol 45: 300-302.
12. Kemmett D, Hunter JAA (1990) Sweet's syndrome: a clinicopathologic review of twenty-nine cases. J Am Acad Dermatol 23: 503-507.
13. Fett DL, Gibson LE, Su WPD (1995) Sweet's syndrome: systemic signs and symptoms and associated disorders. Mayo Clin Proc70: 234-240.
14. Florez A, Sanchez-Aguilar D, Roson E, Prieto A, Van den Eyden A, Toribio J (1999) Sweet's syndrome associated with *Salmonella enteritidis* infection. Clin Exp Dermatol 24: 237-242.
15. Chen HH, Hsiao CH, Chiu HC (2004) Successive development of cutaneous polyarteritis nodosa, leucocytoclastic vasculitis and Sweet's syndrome in a patient with cervical lymphadenitis caused by *Mycobacterium fortuitum*. Br J Dermatol 151: 1096-1100.
16. Singh RK (2002) Acute febrile neutrophilic dermatosis following tuberculous infection. JAPI 50: 1322-1323.

17. Serirat O and Thaipisuttikul Y (2011) Sweet's syndrome associated with *Mycobacterium tuberculosis* and cervical cancer: a case report. J Med Assoc Thai 94 Suppl 2: S119-122.
18. Cohen PR (2009) Neutrophilic dermatoses: a review of current treatment options. Am J Clin Dermatol 10: 301-312.
19. Theng TS, Chan YC, Leow YH, Tan SH (2003) Sweet's syndrome associated with *Mycobacterium chelonae* and herpes simplex virus infections: a case report. Ann Acad Med Singapore 32: 411-414.
20. Rahier JF, Lion L, Dewit O, Lambert M (2005) Regression of Sweet's syndrome associated with Crohn's disease after anti-tumour necrosis factor therapy. Acta Gastroenterol Belg 68: 376-379.
21. Foster EN, Nguyen KK, Sheikh RA, Prindiville TP (2005) Crohn's disease associated with Sweet's syndrome and Sjogren's syndrome treated with infliximab. Clin Dev Immunol 12: 145-149.

Corresponding author
Partha Sarathi Karmakar
Associate Professor
Department of Medicine
North Bengal Medical College and Hospital
Siliguri, West Bengal
India
Email: parthamed@yahoo.co.in

Conflict of interests: No conflict of interests is declared.