Case Report

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


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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


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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.
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.

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
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


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