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Case report

Recurrent non-healing perianal ulcer: A case of tuberculosis cutis orificialis

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Abstract

Tuberculosis cutis orificialis (TCO) is a rare manifestation of cutaneous tuberculosis. Due to its variable clinical features, the diagnosis may be missed at the onset of the disease. Here we describe, TCO as well as intestinal tuberculosis in a patient with recurrent non-healing perianal ulcer. First TCO was detected, later on intestinal T.B was also detected. The perianal lesions appeared as multiple outbreaks of ulcer which resolved totally after combined anti T.B treatment. The clinical course of intestinal lesions and tuberculosis cutis orificialis appeared interrelated.

Keywords: Tuberculosis cutis orificialis; Cutaneous tuberculosis; Perianal ulcer; Intestinal tuberculosis

1. Introduction

Tuberculosis still poses a global threat due to its various forms of presentation. Extrapulmonary T.B has increased in recent years (about 5% of all cases) displaying a wide spectrum of its clinical manifestations, the perianal localization still is rare (0.7%) according to available published data (Alvarez Conde et al., 1992; Candela et al., 1999; Bravo and Gotuzz, 2007). Diagnosis and treatment can be delayed in this type of tuberculosis as the differential diagnosis includes a large spectrum of diseases. Clinically, perianal T.B has been classified by Altinoz et al. (2003) into four subtypes: ulcerative, verrucous, lupoid and miliary. Ulcerative type is the most common type and usually occurring secondary to a focus in the lungs or intestine. The presented case displayed the features of ulcerative periorificial T.B. It began as a small, round nodule, reddish-brown in colour and soft in consistency. Gradually an ulcer with undermined borders with seropurulent discharge developed in the centre of the nodule. The intestinal lesions were asymptomatic until perianal lesions developed.

2. Case report

The presenting case was of a 45 year old Saudi man complaining of recurrent painful ulcerative lesions in perianal area that had started about 6 yrs back. Initial lesion started as a painful nodule, he sought medical advice in the King Fahad Central Hospital, Jizan and was diagnosed as having perianal sinus and was surgically removed. After one and half year, the same lesion recurred with the same symptoms and was surgically removed in a private hospital in Jeddah. Few months later, again he felt discomfort and a

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small ulcer started to appear at same site, and with time the ulcer was progressing with seropurulent discharge. It was associated with fever, night sweating, loss of weight, loss of appetite, generalized body ache and occasional abdominal pain and diarrhoea. There was no history of joint pain, cough or any previous medical illness. Also there was no history of similar condition in the family. The patient was a heavy smoker and gat chewer.

Physical examination revealed deep ulcer with irregular and undermined border in the perianal area 15 × 10 cm in size with pale granulation and seropurulent discharge (Fig. 1). There were multiple, enlarged, non-tender mobile, lymph nodes in the left inguinal area. Abdominal examination showed firm, mobile, non-tender mass in the right lumbar area. There was no ascites nor organomegaly. Cardiovascular and chest examination revealed no abnormality.

Laboratory examination disclosed high erythrocyte sedimentation rate (80 mm/h; normal 1–20) and low haemoglobin (Hb 10 g/dl; normal 12–16). Liver function, renal function and blood sugar were normal. Serology for sexually transmitted diseases including syphilis, human immunodeficiency virus, hepatitis B, C was negative. PCR for human herpes simplex virus, antinuclear factor, ANCA profile and rheumatoid factors was negative.

Microbiological cultures for bacteria, mycobacteria and fungi both from lesional skin and blood yielded negative result. The tuberculin test was positive (see Fig. 2).

Instrumental analyses, like chest X-ray was normal but recto-colonoscopy showed multiple, nodular fungating circumferential ulcerating masses in the proximal part of ascending colon and caecum. Multiple biopsies were taken from the colon which showed granulomatous reaction. Gastroenterologists put the differential of tuberculosis, Crohn's disease, malignancy, basidiobolomycosis and eosinophilic granuloma.

Abdominal sonography showed a heterogeneous mass measuring 3 × 3 cm with calcification noted in the right iliac fossa mostly arising from the ileocaecal junction.

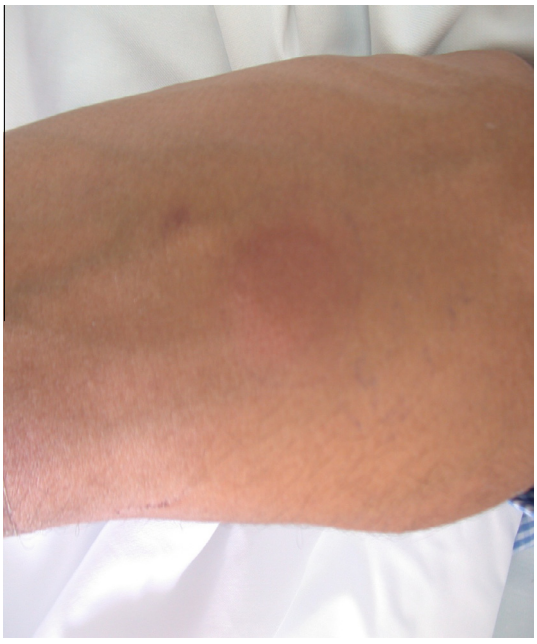

Fig. 2. Tuberculin test showing erythematous, indurated area larger than 15 mm on left forearm.

There was no lymph node enlargement noted in the paraaortic area nor ascites.

C.T abdomen-pelvis showed circumferential mucosal thickening involving about 11 cm of the caecum and ascending colon with pericecal fat stranding and no proximal ileal dilatation or signs of obstruction, a picture of inflammatory-infection process.

Biopsy specimens taken from the edges of the ulcerative lesions showed multiple small caseating granulomas with multinucleated langerhans cells, lymphocytes and histiocytes, a picture consistent with tuberculosis (Fig. 3).

Acid-fast bacilli were seen in the granuloma and necrotic area by Ziehl-Neelsen stain and were rapidly confirmed by polymerase chain reaction (PCR) as mycobacterium tuberculosis.

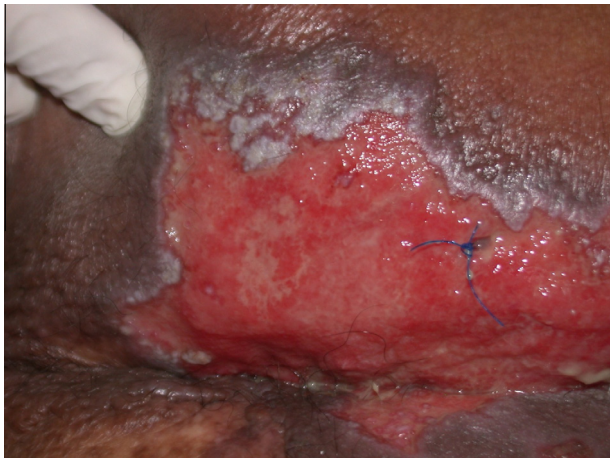

Fig. 1. Showing deep ulcer with irregular, undermined border in perianal area with pale granulation and seropurulent discharge.

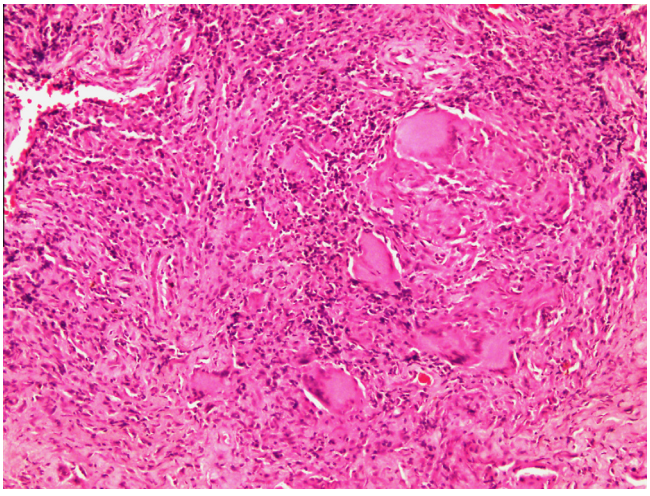

Fig. 3. Showing multiple small caseating granulomas with multinucleated langerhans cells, lymphocytes and histiocytes.

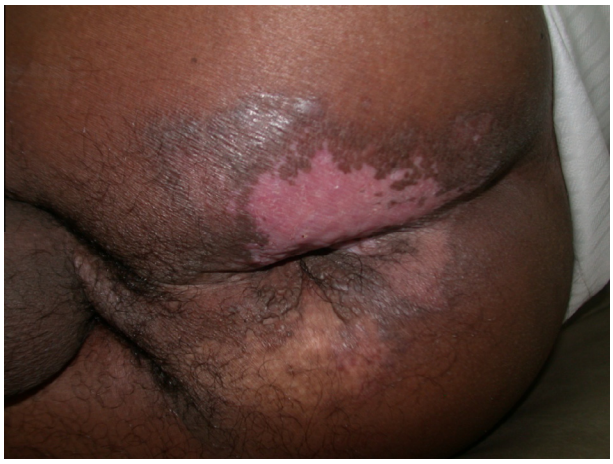

Fig. 4. Showing healed ulcerated lesions with depigmentation.

The patient was started on standard anti-TB therapy, consisting of isoniazid 300 mg, rifampicin 600 mg, pyrazinamide 1000 mg, and ethambutol 900 mg daily for 2 months, followed by an additional 4 months therapy with isoniazid and rifampicin. Daily dressing with silver sulfadiazine cream and silver nitrate gauze was done for 6 weeks. Follow-up after 2 months of initiation of therapy, perianal ulcerations as well as other clinical symptoms completely resolved except some depigmentation in the healed ulcerated area (Fig 4). Six months later, the patient's local and general condition remained unremarkable.

3. Discussion

Skin involvement is a rare form of extrapulmonary tuberculosis, whereas perianal tuberculosis representing a form of cutaneous tuberculosis is even more rare. Tuberculosis cutis orificialis usually results from autoinoculation of the infectious agent in patients with advanced internal tuberculosis of the lungs, gastrointestinal or genitourinary tract (Leon- Mateos et al., 2005; Ghosh et al., 2009). Haematogenous or lymphatic dissemination from another active source of tuberculosis has also been described (Miteva and Bardarov, 2002). Also sometimes as part of disseminated T.B, miliary lesions around anus can be started. Our patient had visceral tuberculosis involving ascending colon and caecum, and perianal involvement was secondary event either by auto-inoculation or haematogenous spread. The bacilli are thought to reach and attack the traumatized perianal mucosa or skin.

The initial manifestation of intestinal tuberculosis can be perianal ulceration as observed in our case. In our patient, there was irregular ulcer with undermined edges and soft consistency on the left perianal area. The differential diagnosis consists of inflammatory bowel diseases like Crohn's disease, deep mycoses, amoebiasis, neoplasia, pyoderma gangrenosum, sarcoidosis and infections with HSV, CMV, HIV, varicella zoster virus (Ozarmagan et al., 2010).

Intestinal tuberculosis and Crohn's disease are similar granulomatous disorders. The clinical, morphological and histopathological features of both diseases overlap to such an extent that it can be difficult to distinguish them (Makharia et al., 2007). Initially this happened to our patient. But later, acid-fast bacilli (AFB) were detected in the granuloma by Ziehl- Neelsen stain and was rapidly confirmed by polymerase chain reaction (PCR) as mycobacterium tuberculosis.

Diagnosis of T.B, especially cutaneous T.B, by conventional laboratory methods is unreliable and time consuming. In one study, PCR produced the best detection rate (79.4%), followed by histopathology (73.5%), various cultures (29–47%) and smear examination (5.8%) (Negi et al., 2005). Based on this, the negative culture in our patient is not unexpected.

Finally, perianal ulcers can be the initial manifestation of tuberculosis in asymptomatic healthy patients. In order to make an early, exact diagnosis and start an appropriate treatment, in addition to histopathological examination, AFB must be searched with Ziehl–Neelsen stain, culture and polymerase chain reaction.

Conflict of interest

None declared.

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