

Received: 11.09.2018

Accepted: 26.01.2019

Published: 31.07.2019

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Miliary tuberculosis presenting as scrofula in a healthy man: a diagnostic challenge

Gruźlica prosówkowa objawiająca się jako skrofuloza u zdrowego mężczyzny – wyzwanie diagnostyczne

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Abstract

Tuberculous lymphadenitis constitutes up to 40% of extrapulmonary tuberculosis. Chronic cervical lymphadenopathy may be the only manifestation of extrapulmonary tuberculosis in the head and neck region. Symptoms, clinical findings and prognosis differ greatly depending on the host's immune system, existing comorbidities, site and extension of infection as well as early diagnosis and commencement of treatment. We report a case of miliary tuberculosis with both pulmonary and extrapulmonary involvement in a healthy male who was successfully treated with surgical drainage of neck abscess followed by 9 months of an anti-tuberculosis drugs treatment.

Keywords: tuberculosis, scrofula, lymphadenitis, lymph nodes, neck

Streszczenie

Gruźlicze zapalenie węzłów chłonnych stanowi do 40% przypadków gruźlicy pozapłucnej. Przewlekła limfadenopatia szyjna może być jedynym objawem gruźlicy pozapłucnej w okolicy głowy i szyi. Objawy, wyniki kliniczne i rokowanie różnią się znacznie w zależności od układu odpornościowego pacjenta, chorób współistniejących, miejsca zakażenia i jego rozległości. Duże znaczenie mają także wczesna diagnoza i szybkie wdrożenie leczenia. Opisujemy przypadek gruźlicy prosówkowej z zajęciem płuc i pozapłucnym u pozornie zdrowego mężczyzny, którego terapia obejmowała chirurgiczny drenaż ropnia szyi oraz stosowanie przez 9 miesięcy leków przeciwgruźliczych.

Słowa kluczowe: gruźlica, skrofuloza, zapalenie węzłów chłonnych, węzły chłonne, szyja

INTRODUCTION

Tuberculous (TB) cervical lymphadenitis, also known as scrofula or King's evil, refers to the infection of cervical lymph nodes by *Mycobacterium tuberculosis* and has been well documented for centuries⁽¹⁾. Typical findings include unilateral, multiple, matted and non-tender cervical lymph nodes^(2,3). Scrofula is often associated with the affiliation to a lower socioeconomic group and immunocompromised patients^(2,4). High index of suspicion is necessary in the diagnosis as a confirmatory result through fine needle aspiration cytology (FNAC) of lymph nodes may be inconclusive at times^(2,3). Therefore, it remains a challenge to diagnose TB lymphadenitis due to varied presentation that may mimic other pathology, and inconsistent laboratory results.

CASE REPORT

A 30-year-old healthy male presented with 3 months history of a painless, progressive, left neck swelling. The swelling was not associated with trauma or an insect bite. He denied odynophagia, dysphagia, shortness of breath, cough, haemoptysis, hoarseness and constitutional symptoms. He had no history of smoking, alcohol consumption or high-risk behaviours such as drug abuse or sexual promiscuity. However, he stayed with a close relative who was diagnosed and treated for pulmonary TB for 7 years. On the neck examination, a soft, well-circumscribed, cystic, non-tender swelling with the size of 5 × 5 cm was palpated over the left neck, just above the left clavicle. There was no other palpable cervical lymph node. Other examinations were unremarkable. Full blood count, renal profile, liver function test, erythrocyte sedimentation rate (ESR) were within normal range. Serology for hepatitis B, C, human immunodeficiency virus (HIV), syphilis and TB workout such as chest radiograph (CXR), sputum

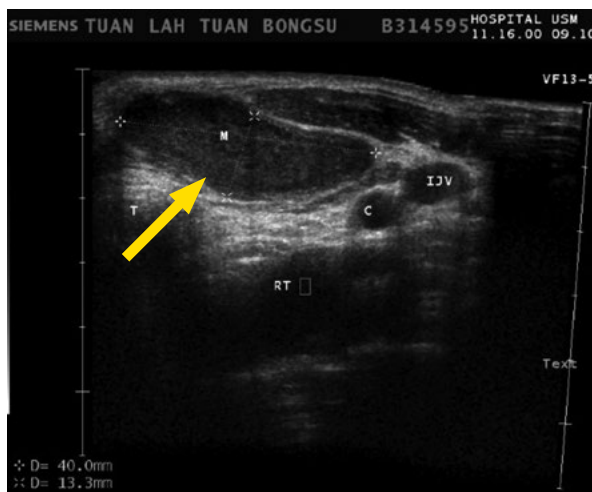


Fig. 1. A well-defined, oval, homogenous mass with the size of 4.0 × 1.3 × 3.5 cm (yellow arrow) in between the left thyroid lobe and left SCM muscle likely to be cervical lymphadenopathy

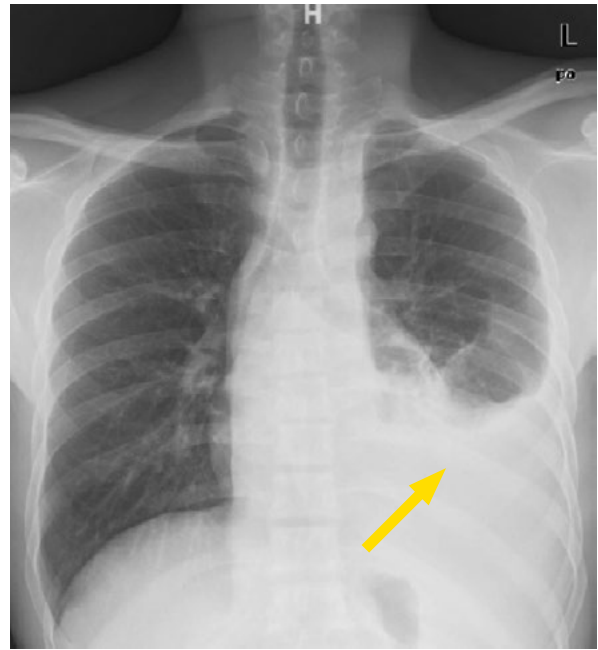


Fig. 2. A repeated CXR showed unilateral pleural effusion over the left lung that necessitates further investigation for evaluation such as CECT thorax

for acid-fast bacilli (AFB) and Mantoux test were all negative. Ultrasonography (USG) of the neck showed a single, homogenous mass that might represent a benign nodule such as a nodal enlargement over the left sternocleidomastoid (SCM) muscle (Fig. 1). FNAC was reported as compatible with an abscess. Incision and drainage were performed, and 30 mL of thick pus was evacuated. The pus was sent for bacterial culture and AFB stain, which turned out to be of no growth and negative, respectively. The patient was then discharged home with oral amoxicillin/potassium clavulanate, and daily dressing at the nearest clinic.

At a 2-month follow up, his wound remained open with a minimal discharge despite an adequate oral antibiotic coverage and dressing. A re-exploration and wound debridement were performed. Intraoperatively, granulation tissues with cheesy materials were removed and sent to the lab. The histopathological examination (HPE) of the tissue was reported to be granulomatous inflammation. No AFB or fungal bodies were seen after using Ziehl-Neelsen (ZN) and Gomori methenamine silver (GMS) special stains respectively. One month later, the patient presented with mild shortness of breath, chesty cough with whitish sputum. A repeated CXR revealed pleural effusion over the left lower zone (Fig. 2). A contrast-enhanced computer tomography (CECT) scan of the neck, thorax and abdomen was ordered for further evaluation. There were multiple collections seen over bilateral paraspinal muscles, the right psoas major muscle and the left SCM as well as a massive left pleural effusion (Figs. 3–6). In correlation with clinical features and CECT findings, a diagnosis of miliary TB was made and patient was started on empirical anti-TB medications. The patient was prescribed

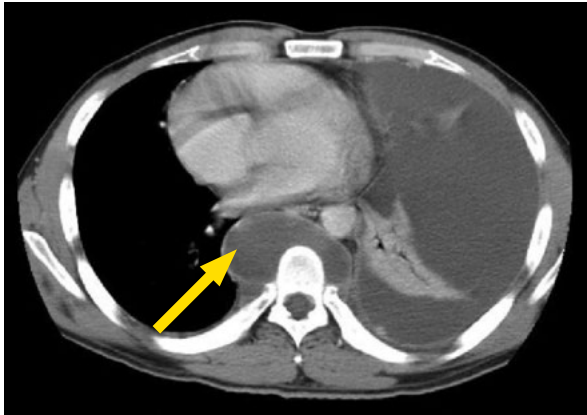


Fig. 3. Axial view of CECT thorax showing massive left pleural effusion with bilateral paraspinal collection, over the right side (yellow arrow)

1 g streptomycin, 300 mg isoniazid, 600 mg rifampicin and 2 g pyrazinamide (SHRZ regimen) daily for a total of 9 months. The productive cough resolved, neck wound closed up spontaneously after 2 weeks and by 1 year, the patient was well and discharged from the follow up.

DISCUSSION

Scrofula was historically derived from a Latin word that carries the meaning of brood sow⁽¹⁾. Centuries ago, many believed that the kings of England and France possessed a supernatural power to heal scrofula, just by touching



Fig. 4. Coronal view of CECT thorax and upper abdomen showing massive left pleural effusion and bilateral paraspinal collection, extending from superior mediastinum (level T1) until lumbar region (level L1)

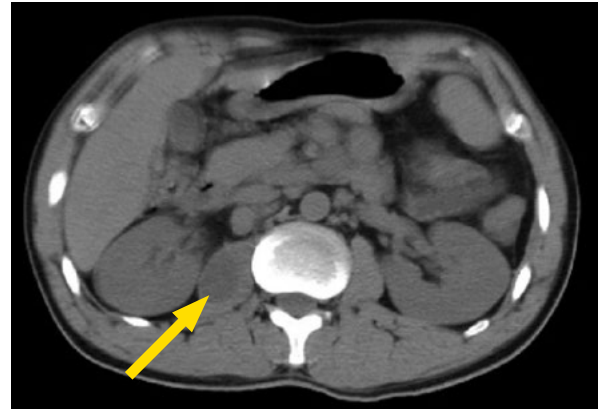


Fig. 5. Collection extend downwards into the right psoas major (yellow arrow). The left psoas major is not involved

the sufferers⁽¹⁾. The mean age of occurrence is 25.6 years old, with no sexual predilection⁽²⁾. Paediatric and elderly groups are more prone to contract TB due to the immaturity or waning of their immune systems^(3,5). Poor nutrition and living condition may increase the risk of exposure to TB⁽²⁾, like in our patient who shared a small house with a pulmonary TB patient without proper preventive measures. Other immunocompromised groups include post-splenectomy patients, intravenous drug abusers and HIV seropositive patients⁽⁴⁻⁶⁾.

TB cervical lymphadenitis typically present with painless neck swelling of weeks to months duration⁽⁴⁾. In a study of TB cervical lymphadenitis in 65 patients, most were found to present unilateral (87.7%), multiple (82.3%) and matted (68.6%) lymph nodes⁽²⁾. The most common involved lymph nodes were at level V (59.4%) and level II (42.2%)⁽²⁾. About 21.5% of these cases presented with abscess, while another

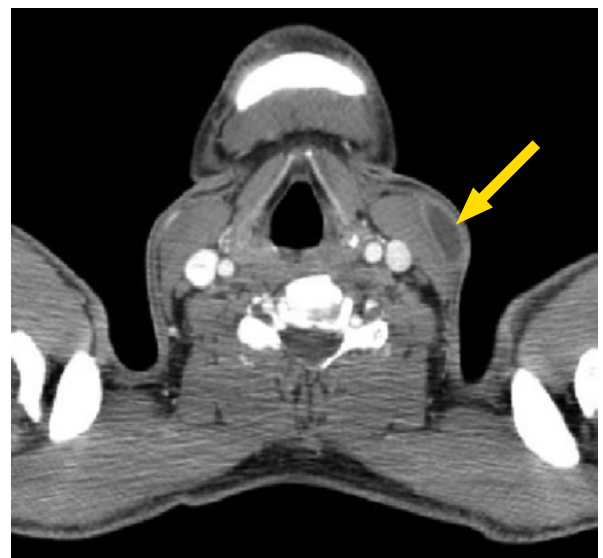


Fig. 6. Axial view of CECT neck showed intramuscular collection size of 2.1 × 0.9 × 6.0 cm within the left SCM muscle (yellow arrow)

9.2% had discharging sinus⁽²⁾. In a different study that involved 893 patients with scrofula, 18% had associated pulmonary TB⁽³⁾. Systemic symptoms such as fever, poor appetite, weight loss, fatigue and night sweats may be present only in immunocompromised patients⁽⁴⁾.

TB is an infectious airborne disease. Once the mycobacterium reaches the alveoli, it has 3 potential fates depending on the host's immune system⁽⁷⁾. The mycobacterium can either be destroyed by the immune system and the patient gains immunity (first scenario), or it can multiply and cause pulmonary TB (second scenario), or it becomes dormant and stop progressing until the patient develops the reactivation of TB which involves the impairment of immune system like malnutrition, malignancy or HIV (third scenario). During a primary pulmonary TB infection, the mycobacterium can travel to other parts of the body through hematogenous or lymphogenous dissemination, stay concentrated at particular sites and lie dormant for years before causing disease⁽⁷⁾. Our case most likely belongs to the group in the third scenario because the patient had frequent contact with a pulmonary TB patient and the clinical symptoms appear only after 7 years of the dormant stage. Diagnosis in this patient was a challenge because of negative results from FNAC, HPE and TB workout. FNAC has 83.9% accuracy in diagnosing TB and can be further enhanced by the adjunctive usage of ZN stain^(2,8). Massive left pleural effusion with multiple intramuscular cold abscesses in CECT coupled with previous TB exposure were adequate evidences to start anti-TB treatment. According to the World Health Organization (WHO) 2006 guideline for smear negative pulmonary TB, empirical anti-TB therapy can be initiated based on CXR and the clinician's judgement⁽⁹⁾. Initiation of anti-TB drugs in extrapulmonary TB without positive culture is justified when there is a difficulty in obtaining good sample from anatomical challenging sites or from critically ill patients⁽⁹⁾. WHO recommends continuous monitoring in such cases because progressive clinical improvement equate to correct diagnosis of TB⁽⁹⁾. Medical therapy with anti-TB drugs is the most favourable choice for pulmonary, extrapulmonary and disseminative TB with a very good outcome⁽³⁾. Surgical approach is reserved for selected refractory cases that respond poorly to the medical therapy⁽³⁾.

CONCLUSION

In the ideal situation, the diagnosis of TB cervical lymphadenitis is made with positive FNAC or HPE results. Sometimes, the laboratory results may be inconclusive to suggest TB despite repeated biopsies. Therefore, imaging modality such as CECT can be included to the diagnostic arsenal. In this case, despite the administration of adequate antibiotic drugs, surgical drainage of abscess and re-exploration, the patient still came back to the clinic with gaping neck wound and recurrent discharge after 2 months of operation. Clinical presentation like this should prompt clinicians on the possibility of TB.

Conflict of interest

The authors do not declare any financial or personal links to other persons or organizations that could adversely affect the content of this publication and/or claim rights thereto.

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