Case report

A very rare case of scrofuloderma with multiple cervical lymphadenitis tuberculosis

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ABSTRACT

Scrofuloderma is a clinical form of secondary cutaneous tuberculosis. It is commonly characterized by bluish-red nodules that cover the lymph nodes, bones or joints, disrupting the affected tissue to be replaced with granulation tissue. We reported a case of 10-year-old boy with ulcers in the cervical and axillary region. The lesion began as papules, which gradually developed into nodules and pustules. Chronic cough was also found, however chest x ray was normal and sputum direct smear for acid fast bacil was negative. Histopathologic finding of tissue biopsy revealed epidermal necrosis in the central part surrounded by granulomatous tissue forming an abscess and histiocyte infiltrates, confirming the diagnosis of scrofuloderma. CT Scan of neck showed multiple enlarged lymph nodes, and FNAB result was suggestive for tuberculous lymphadenitis. Patient was then given anti-tuberculosis therapy. Four months later, the ulcers began to resolve and heal gradually.

1. Introduction

Tuberculosis has been found since prehistoric times and to date is still one of infectious disease with high burden of mortality [1]. The WHO Global Tuberculosis Report 2016 stated Indonesia as the second highest country with TB prevalence around the world [2]. The Global Burden of Disease study reported TB as the second leading cause of death in the world. Tuberculous lymphadenitis is the most common extra pulmonary tuberculosis, on the other hand, cutaneous tuberculosis is a very rare case accounting for 1–2% of all tuberculosis cases [3].

Scrofuloderma and verrucous cutis tuberculosis is a clinical form of secondary cutaneous tuberculosis caused by Mycobacterium tuberculosis and atypical Mycobacterial. Scrofuloderma occurs due to direct spread from organisms under the skin that have been infected by M. tuberculosis bacilli, particularly lymph nodes, but can also come from joints, tendons, synovial fluid and bones. Chronic and severe inflammation will lead to rupture and ulceration of the lymph node. Suppurative ulcers are frequently found in cases of cutaneous tuberculosis. Regions most commonly affected include chest, neck and axillary lymph node [4].

Clinical features of scrofuloderma begins with multiple or solitary painless lymph node enlargement. Over time, the bottom part will become softened creating cold abscess. The abscesses will become suppurated and eventually rupture, forming a linear and irregular ulcer with bluish red (livide) surface around it. The differential diagnosis of verrucous cutaneous tuberculosis include atypical mycobacterial infection (NTM), sarcoidosis, verrucous vulgaris, blastomycosis, leprosy, and tertiary syphilis. Treatment of scrofuloderma is similar to the treatment of pulmonary tuberculosis by using oral anti-tuberculosis regimen containing isoniazid, rifampicin, pyrazinamide, ethambutol [5].

2. Case presentation

A 10-year-old boy came with ulcer in neck and axilla region. The lesion began as papules that gradually developed into nodules, pustules and painless suppurative ulcer (Fig. 1A). Patient also had chronic cough with yellowish sputum since several months along with low grade fever and weight loss.

Physical examination revealed ulcer with suppurative surface in the bottom part, along with nodule measuring 4 cm x 0.5 cm (Fig. 1A). Around his ear, some crusts were found measuring 3 cm x 0.5 cm typical for scrofuloderma (Fig. 1B). In axillary region, there were also painless ulcers with pustulated sinus and purulent discharge, surrounded by bluish and edematous skin surface (Fig. 1C). There was also mobile and painless lymph node enlargement in the left supraclavicular...
region measuring 2 × 2 cm.

Chest X-ray was within normal limit. Tuberculin skin test was positive with 10mm induration. However, direct smear of sputum specimen was negative for acid fast bacilli. Excisional biopsy of the ulcers was performed and the histopathology result showed epidermal necrosis in the central region surrounded by granulomatous tissue peripherally, forming abscess with histiocytic infiltrate around it, confirming the diagnosis of scrofuloderma.

He underwent CT Scan of the cervical region and multiple lymph node enlargement was found, suggestive for tuberculous lymphadenitis (Fig. 2A and B). Thus, we did FNAB on the lump and the cytology findings showed groups of epitheloid cell with spindle nucleous with chromatin granules spread equally, surrounded with basophilic amorf mass and fibrous tissue. The background staining of the smear was mature lymphocyte, lymphoblast, neutrophil and macrophage, confirming the diagnosis of tuberculous lymphadenitis (Fig. 2C). He was then diagnosed with scrofuloderma with multiple cervical tuberculous lymphadenitis.

He was treated with standard regimen of anti-tuberculosis drugs containing Rifampicin (R), Isoniazid (H), Pyrazinamid (Z), and Ethambutol (E) for 2 months continued by Rifampicin (R), Isoniazid (H) for the next 4 months. In the fourth months, the lymph node enlargement in the cervical region were shrinked (Fig. 3A and B). The ulcers in the neck and axilla began to resolve gradually leaving scar tissue (Fig. 3C). In some spots, some crusts were still found. Overall, he showed a satisfying response to the anti-tuberculosis therapy.

3. Discussion

The prevalence of tuberculosis is estimated to reach 9.6 million case, in which 1 million cases were found in children. Diagnosis of cutaneous tuberculosis is quite challenging for clinician because it has similarities with other skin lesions and many other cutaneous clinical manifestations. Cutaneous tuberculosis was firstly described in 1981 by Beyt et al. [6].

The most common form of cutaneous tuberculosis is scrofuloderma, particularly found in children. Scrofula is an old term referring for tuberculosis infection of lymph nodes in the neck, known as cervical tuberculosis lymphadenopathy. It is generally the result of primary tuberculosis infection of the lymph node. Mycobacterium tuberculosis bacilli can spread through lymph node and hematogenously. Scrofuloderma refers to a condition characterized by a bluish-red nodule overlying an infected lymph gland, bone or joint that breaks down to form an undermined ulcer with a granulating tissue at the base. Progression of the disease leads to irregular adherent masses, densely fibrous at some spots while fluctuant and discharging at others [4,5,7].

The diagnosis of scrofuloderma is mostly confirmed by histopathological finding of tissue biopsy especially in developing countries. The presence of acid-fast bacilli or caseating granulomatous tissue with langhans cell make a definite diagnosis, just like the findings in this patient. However, some studies found the diagnostic performance of Xpert MTB/RIF, conventional polymerase chain reaction (PCR), mycobacteria growth indicator tube (MGIT). Gautam et al. found that the detection rates of Mycobacterium tuberculosis complex (MTBC) by Xpert MTB/RIF, conventional PCR, and MGIT were 26%, 21%, and 18%, respectively. Both the tests of Xpert MTB/RIF and PCR, PCR and MGIT, Xpert MTB/RIF and MGIT were positive in 16%, 16%, and 11% of cases, respectively. Individual modalities of the diagnosis are already available, but all have drawbacks with varied sensitivity and specificity. Combined using the available clinical, radiological, and microbiological modality to reach early diagnosis can go a long way to avoid misdiagnosis and unnecessary delay in treatment, especially in cases, without the pulmonary involvement [8].

World Health Organization recommendation for cutaneous tuberculosis is anti-tuberculosis regimen containing Rifampicin (R), Isoniazid (H), Pyrazinamid (Z), Ethambutol (E) for 2 months continued by Rifampicin (R), Isoniazid (H) for the next 4 months. In severe case, surgical approach like electrosurgery, cryosurgery, and curettage by electrodiication may sometimes be needed [9].
4. Conclusion

We reported a very rare case of scrofuloderma along with multiple cervical lymphadenitis. This form of cutaneous tuberculosis is quite challenging to diagnose due to its similarity to many other skin lesions, and thus, multidisciplinary approach is necessary in order to establish a definite diagnosis. After being treated with anti-tuberculosis drugs, the ulcers began to resolve and heal gradually.

Consent for publication

The father of patient was properly informed and had provided consent for the clinical information of his son to be included in the publication of this case report and the accompanying images.

Conflicts of interest

The authors declare that they have no competing interests.

Authors’ contributions

NNS described and designed the article. NNS, EGH, and AY were involved in treating the patient. NNS, EGH and AY participated in editing the manuscript critically. All authors declared that they contributed to this article and that they have read and approved the final manuscript.

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Appendix A. Supplementary data

Supplementary data related to this article can be found at https://doi.org/10.1016/j.rmcr.2019.100842.

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Fig. 3. (A&B). Four months later, the lymph node enlargement in supraclavicular region was getting smaller, ulcers in the neck began to heal. (C) Ulcers in the axillary region resolved leaving scar tissue.