Tuberculous Dactylitis in a Case of Multiple Scrofuloderma

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Abstract
This paper reports a case of tuberculous dactylitis and multiple scrofuloderma spreading through the lymph nodes. Scrofuloderma, also known as tuberculosis colliquativa cutis, is a form of cutaneous tuberculosis (TB) that occurs most often in children and young adults and involves the skin over the infection focus (i.e., lymph nodes, bones, or joints). Scrofuloderma can affect the lower limbs and upper arms by spreading osteomyelitis TB on the humerus, wrist, and elbow. This study reports the case of a 19-year-old man who initially developed painful and swollen skin, followed by the appearance of numerous recurring lumps on the left arm and hand and the right foot, as well as the folding right hamstring, over 3 years. The patient had no clinical improvement with antibiotics and excision. Radiography of the left hand showed tuberculous dactylitis. A biopsy of the left arm was performed, and Ziehl-Neelsen staining showed acid-fast bacilli. Mycobacterium tuberculosis was confirmed by a real-time polymerase chain reaction. Anti-TB drug treatment was initiated with rifampicin, isoniazid, pyrazinamide, and ethambutol, and debridement was performed on the left hand, which resulted in significant improvement of the lesion. Atypical clinical manifestations and unawareness of M. tuberculosis as an underlying disease delayed the diagnosis and treatment of this patient with tuberculous dactylitis and multiple scrofuloderma.
Introduction

Tuberculosis (TB) is an infectious disease caused by *Mycobacterium tuberculosis* and is one of the ten leading causes of mortality worldwide. After India and China, Indonesia is the third country with the highest number of TB cases. In 2018, the number of TB cases was 10 million globally, with 1.2 million mortalities. As many as 44% of TB cases occur in Southeast Asia [1]. Most cases were pulmonary TB, whereas fewer than 20% of cases were extrapulmonary TB [2].

Scrofuloderma, also known as *tuberculosis colliquativa cutis*, is a form of cutaneous TB that occurs most often in children and young adults and involves the skin over the infection focusing on lymph nodes, bones, or joints. It can initially be a hard and painless subcutaneous nodule, gradually enlarging, suppurating, and then ulcerating. Scrofuloderma is mostly found on the neck (parotid, submandibular, and supraclavicular), axilla, and inguinal areas [3, 4]. It rarely occurs on the lower legs or upper arms by the spread of osteomyelitis of the humerus, wrist, and elbow bones [5, 6].

The diagnosis of cutaneous TB is established by the patient’s medical history, risk factors, physical examination, and supporting examinations, including histopathologic examination, specimen culture, polymerase chain reaction (PCR), interferon-gamma release assay, and the tuberculin skin test [3, 4, 7]. The World Health Organization recommends a two-phase plan for cutaneous or extrapulmonary TB treatment regimen. First, rifampicin, isoniazid, pyrazinamide, and ethambutol are administered for 8 weeks in the intensive phase; second, rifampicin and isoniazid are administered until 2 months in the continuation phase after the lesions heal [3, 7, 8]. This report presents a rare presentation case of multiple scrofuloderma and tuberculous dactylitis.

Case Report

A 19-year-old man presented to the Tropical Skin Infection Division, Dermatology and Venereology Outpatient Clinic, Dr. Cipto Mangunkusumo National General Hospital, Jakarta, Indonesia, because of a 3-year history of lumps and unhealed wounds on the left hand and the right foot. Initially, the skin was swollen and sore, and then lumps appeared, disintegrated, and became scars. Another lump resurfaced after the scar dried, followed by unhealed wounds surrounding the previously affected areas. The patient described the lumps as feeling hard and sore and reported that they secreted a yellowish liquid. The lumps were often not accompanied by pain. Similar lesions appeared on the left upper extremities and then spread to the left axilla in the previous year. A new similar lesion appeared on the right hamstring 4 months ago. This lesion also began as a lump, which then ruptured and formed a scar. The patient denied any history of trauma before the appearance of the lumps and any personal TB history or TB in his family and closest friends. The father (working as a delivery courier), mother, and two younger siblings reside with the patient. In addition, the patient received the Bacille Calmette-Guérin (BCG) vaccine during childhood.

The patient previously sought evaluation and treatment 3 years ago for the lesion on the left hand and right foot. The diagnosis was not known at this time. Moreover, antibiotics were given and surgery was performed to remove the lump but the patient did not experience improvement. An antibiotic, the name of which was unknown to the patient, was prescribed and the patient adhered to the drug therapy for 1 year. Following the patient’s preference, the wound was treated at a wound care clinic although the surgery was performed at a hospital.

At the current presentation, the patient reported a decreased appetite and weight loss of 12 kg in the previous 3 years, in addition to intermittent recurring fever. The patient had
normal vital signs and body mass index. Physical examination revealed multiple ulcers on the axilla and medial left arm, with five ulcers. Their appearance was irregular, with granulation tissues; some ulcer walls resonated; the edges not raised but were crusted; the surrounding tissue appeared erythematous to livid; and warmth was not palpable. On a visual analog scale, the patient rated his pain score as 1–2. Hypertrophic and eutrophic scars, which were bluish and erythematous, multiple, irregularly shaped, and linearly arranged, were noted on the patient’s left upper arm, left hand, and right foot.

Physical examination of the posterior right upper leg showed a solitary nodule with an erythematous surface, without fluctuation, and felt solid and warm on palpation. The patient’s pain rating on a visual analog scale was 3–4. Examination of the lymph nodes revealed an enlargement of the left axilla and right lateral inguinal nodes. One enlarged lymph node on the left axilla had a 0.5-cm diameter with a rubbery consistency, without pain elicited on touching and with detectable matting. In addition, two lymph node enlargements in the right lateral inguinal area were found, with a diameter of 0.5 and 1 cm, and each with a rubbery consistency, without pain elicited on touching and with detectable matting. Clinical features before the treatment are shown in Figure 1.

Histopathologic examination of the skin lesion on the left hand revealed granuloma with inflammatory cells, lymphocytes, polymorphonuclear leukocytes, and plasma cells. Results of real-time PCR for *M. tuberculosis* were positive, and acid-fast bacilli were shown on Ziehl-Neelsen staining. Results were negative on the Lowenstein-Jensen medium culture, testing for fungi, and aerobic bacteria tissue culture. No radiologic abnormalities were found in the heart, lungs, humerus sinistra, genu dextra, and femur dextra. However, radiologic imaging of the manus sinistra showed a blastic lesion with cortical erosions and irregularities in metacarpal I and soft tissue swelling, which was suspicious for a tuberculous dactylitis (Fig. 2). In addition, radiologic imaging of the pedis dextra showed destructive lytic lesions on the anterior OS calcaneus and inferior OS cuboid with soft tissue swelling, suggestive of osteomyelitis. Based on these findings, the patient was diagnosed with multiple scrofuloderma and TB dactylitis. The scrofuloderma diagnosis was based on the results of a skin biopsy and a PCR positive for *M. tuberculosis*.

Pharmacologic treatment was initiated as a two-phase regimen of oral anti-TB drugs for 9 months. The drugs in the intensive phase during the first 2 months were rifampicin (450 mg), isoniazid (225 mg), pyrazinamide (1,200 mg), and ethambutol (825 mg), per daily. After this phase, the wounds on the previous lumps had dried and properly closed, and no new lumps and wounds resurfaced. The patient reported resolution of pain, fever, and skin irritation. Debridement was performed on the manus sinistra region by an orthopedic surgeon. In the continuation phase during the next 7 months, the pyrazinamide and ethambutol mg were stopped, but the other two drugs were continued at lower doses with rifampicin (300 mg) and isoniazid (150 mg). The patient reported resolved lesions of the lumps, without new wounds and without pain or swelling. The wounds left irregular hypertrophic scars. Clinical features after treatment are shown in Figure 3.

**Discussion**

This patient was diagnosed with scrofuloderma based on medical history and results of physical, laboratory, and imaging examinations, including histopathology, PCR, and radiology. Scrofuloderma is a multibacillary cutaneous TB variant affecting people with weak immune systems [9]. This patient was 16 years old when the lumps on the left hand and right foot initially developed. Scrofuloderma mainly affects children, adolescents, and older adults [3]. In this case, disease onset occurred during adolescence; however, the patient’s initial evaluation
and treatment plan did not immediately recognize the condition as a mycobacterial infection, and the patient often treated himself without any physician’s recommendation. Other contributing factors to cutaneous TB include living in poverty [9].

Additional medical history supporting this patient’s diagnosis was illness >3 years that was unresponsive to antibiotics or invasive treatment. The patient’s lumps and pain recurred surrounding the lumps that had been surgically removed. Scrofuloderma is a type of cutaneous TB for which the route of transmission is endogenous due to direct per continuitatem spread from the organ with TB infection, such that the disease continues to emerge from the underlying organ when a lump is removed, which manifests a new lump around the area of the previously removed lump [9]. Nevertheless, spontaneous healing may occur, although it can take years before scar tissues thoroughly supplant the lesion [3].

The organs underlying the skin that can cause scrofuloderma are the lymph nodes, bones, and joints [3, 4, 7]. In this patient, lumps were found in the axilla to the medial left arm.
The axillary region is vulnerable to scrofuloderma, starting with lymphadenitis in several glands and then increasing in number and becoming more confluent. The initial lesion was a subcutaneous nodule that ruptured and formed ulcers. The appearance of a scrofuloderma ulcer is typically elongated and irregular in shape, with livid surrounding tissue, resonant walls, and granulation tissue covered with seropurulent or caseous pus highly consisting of *M. tuberculosis* [8, 10]. This distinct clinical picture was found in the axillary region to the patient’s left upper arm, medial side.

The patient’s lesions on the left arm to the left hand and the right foot also appeared hypertrophic with eutrophic scars, which, during the disease course, were preceded by lumps that then broke up, after which scars formed. Radiographic examination of the left manus showed TB dactylitis, a manifestation of TB infection in the short tubular bones (e.g., the phalanges, metacarpals, and metatarsals) [11]. Additional examination with magnetic resonance imaging of
bone and culture from the bone biopsy tissue is the standard for diagnosing TB dactylitis. However, TB musculoskeletal infection can also be established if mycobacteria are found in other locations and appropriate clinical manifestations, with or without histopathology, exist [12].

An erythematous nodule breaking into an ulcer, with the surrounding tissue appearing livid, was observed on the patient’s posterior right upper leg. Radiographic examination of the right femur and genu indicated no abnormalities leading to bone damage caused by TB infection. The pathologic mechanism has not been fully elucidated for this lesion. The possibility of spread through the enlarged lymph nodes in the right inguinal or primary inoculation in the skin cannot be ruled out. Consequently, the ulcer improved to a hypertrophic scar after 9 months of treatment.

**Fig. 3.** Clinical features on the skin after anti-TB drugs for 9 months. **a** Post-therapy hypertrophic scars on axilla and left arm. **b** Post-therapy hypertrophic scars on left hand. **c** Post-therapy hypertrophic scars on right upper leg.
A skin biopsy with suspected cutaneous TB was performed 3 years after the initial presentation. The biopsy and PCR examination results indicated a diagnosis of cutaneous TB. The PCR test has high sensitivity and specificity at 100% in multibacillary cases and is lower in paucibacillary cases [4, 7]. Culture examination on Lowenstein-Jensen medium did not indicate growth; its sensitivity for TB is only 23% [7].

Cases of TB in short tubular bones (e.g., the phalanges, metacarpals, or metatarsals) usually occur in pediatric patients aged <6 years. In this age group, the hematopoietic bone marrow is a fertile place for hematogenous bacterial implants. Tuberculous dactylitis is extremely rare in adults. A literature search revealed only 2 cases of multiple scrofuloderma with tuberculous dactylitis in children. Both cases affected young children, 7 and 8 years old, and occurred in TB-endemic areas, one of which was India. Both cases reported risk factors – the 7-year-old did not receive the BCG vaccine, and the 8-year-old had malnutrition [13, 14]. The difference between these cases and the current patient is that the current patient first experienced symptoms at 16 years old and had already received the BCG vaccine. Further, in this patient, a 12-kg weight loss occurred over 3 years. Thus, malnutrition should be reviewed as one of the risk factors for TB in patients although the patient’s body mass index was still within normal limits.

The literature search also found 1 adult case, in which a 20-year-old man had multiple scrofuloderma lesions on his hands and feet with osteomyelitis from the bone underneath. The bones in those cases were the first metacarpophalangeal of the right hand, first and fifth metacarpophalangeal of the left hand, and first metatarsophalangeal of the left foot. In that case, similar to the patient of the current study, the diagnosis was delayed for more than 3 years since the initial lesion appeared. A thorough examination was required, including blood tests, skin tissue biopsies, and cultures, as well as radiologic examinations to establish the diagnosis [15]. The patient’s history was notable for an insect bite 6 years before the emergence of the initial lesion on the right hand, and family members had similar experiences. Immunodeficiency states, e.g., human immunodeficiency virus (HIV), were excluded. In contrast, the patient in the current study had no trauma preceding the lesion and no similar conditions were noted in family members. However, the patient in this study did not undergo immunodeficiency testing, and HIV was not ruled out, which is a risk factor for scrofuloderma [16]. The patient will be advised for HIV testing at the next follow-up examination.

The patient in the current study was given oral anti-TB drugs for 9 months and responded well with closed ulcers, decreased lymph node swelling, and no new lumps. The patient underwent sequestrectomy, guttering, and debridement of the lesion after being diagnosed by an orthopedic surgeon with osteomyelitis TB on the first left metacarpal. The recovery criteria for scrofuloderma patients are all fistulas and ulcers closing, lymph nodes shrinking to <1 cm in diameter, solid consistency, nonerythematous cicatrices, and an erythrocyte sedimentation rate decreasing or returning to normal [9]. In the 3 cases in the literature, all patients received anti-TB drugs (rifampicin, isoniazid, pyrazinamide, and ethambutol) for 2 months, followed by rifampicin and isoniazid for 4–5 months. In all cases, treatment provided good results manifested in the covered ulcers and scar formation [13–15].

**Conclusion**

This case illustrates distinct clinical symptomology for TB infection, which is a long period of illness that is unresponsive to antibiotics or invasive surgery. However, the patient’s insufficient knowledge of the condition and poverty led to a lack of treatment at a proper medical facility. This delay caused a TB infection to develop on the skin and bones affecting several regions, including the axilla, upper left arm, and upper right leg. The patient’s adherence to the
current treatment plan substantially contributed to the recovery. However, the causes of this type of TB infection are still in need of further study, with a focus on the patient’s living environment, the presence of malnutrition, and immunodeficiency status (e.g., HIV infection).

**Statement of Ethics**

Ethical approval is not required for this study in accordance with local guidelines. Written informed consent was obtained from the patient for publication of the details of their medical case and any accompanying images.

**Conflict of Interest Statement**

The author has no conflicts of interest to declare.

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**Author Contributions**

Sri Linuwih S.W. Menaldi was in charge of the literature review, construction of the conceptual design, manuscript review, and editing; Farah Faulin Lubis and Jhauharina Rizki Fadhilla were in charge of the literature review, construction of the conceptual design, data extraction, and writing the original manuscript; Sandra Widaty was in charge of construction of the conceptual design, manuscript review and editing, and overall supervision.

**Data Availability Statement**

All data that support the findings of this study are included in this article. Further inquiries can be directed to the corresponding author.

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