Tuberculous retropharyngeal abscess with Pott disease and tuberculous abscess of the chest wall
A case report
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Abstract
Rationale: Tuberculous retropharyngeal abscess is rare, but it can be fatal if not treated appropriately. It usually occurs secondary to tuberculosis of the cervical spine. Moreover, tuberculous abscess involving the chest wall is relatively rare in skeletal tuberculosis. Although the optimal treatment is controversial, most clinicians suggest a combination of sufficient antituberculous medication and complete resection to prevent recurrence and increase therapeutic efficacy. Herein, we present an unusual case of retropharyngeal abscess with cervical Pott disease and tuberculous abscess of the chest wall.

Patient concerns: The patient was a 27-year-old Indonesian woman who had neck pain, dysphagia, and odynophagia, but no neurological deficit. Examination of the oral cavity showed anterior displacement of the posterior pharyngeal wall. The mass over the right anterior chest wall measured approximately 5 × 4 cm in size.

Diagnoses: Radiography and computed tomography findings were suggestive of retropharyngeal abscess extending to the cervical spine and chest wall abscesses.

Interventions: She was admitted to the hospital for treatment. Drainage of the retropharyngeal and chest wall abscesses with debridement of the chest wall was performed.

Outcomes: No complications occurred after early surgical treatment and administration of antituberculous medication. The patient recovered well and went back to her own country after discharge.

Lessons: Tuberculous retropharyngeal abscess with Pott disease and tuberculous abscess of the chest wall are both complicated diagnoses that physicians have to consider in similar patient presentations.

Abbreviations: AFS = acid-fast stain, CT = computed tomography, ENT = ear, nose, and throat, HIV = human immunodeficiency virus, MRI = magnetic resonance imaging, PCR = polymerase chain reaction.

Keywords: antituberculous drug, case report, chest wall, Pott disease, retropharyngeal abscess, tuberculosis

1. Introduction
Chronic retropharyngeal abscess is often caused by tuberculosis and is primarily seen in adults.[1] Odynophagia and dysphagia are the principal symptoms of retropharyngeal abscess, and it is usually associated with a direct extension from adjacent cervical Pott disease.[2] In the United States, the incidence of Pott disease was 0.05 cases per 100,000, and the proportion of Pott disease cases among all tuberculosis cases was 2.6% in 2011.[3] Tubercular involvement of the cervical spine is unusual in Pott disease.[11] The diagnosis of retropharyngeal abscess is, however, difficult due to the nonspecificity of its symptoms. It should be suspected in patients with a retropharyngeal mass and a destructive lesion of the vertebra.[2] Early diagnosis and treatment with antituberculous medication and surgical intervention are important to prevent further complications, such as airway obstruction or neurological deficit.[1,4]

Skeletal tuberculosis rarely develops in all types of tuberculosis; further, chest wall involvement is not common in skeletal tuberculosis.[5] Hematogenous dissemination from activation of a dormant tuberculous focus is the most common pathogenesis of rib tuberculosis.[6–7] Cases of primary chest wall tuberculosis with no other tuberculosis site have also been reported. Diagnosis of abscess of the chest wall may be challenging owing to the difficulty in differentiating them from a pyogenic abscess or chest wall tumor.[8] Soft tissue tumors are the most common type of lumps in the chest wall and are most important in differential diagnosis. The treatment of chest wall abscess remains controversial.[5] Most patients require a combination of medical and surgical management to increase efficacy and to prevent recurrence.[8] Two retrospective studies reported a recurrence rate after combination therapy of 7.8% and 15%.[8,9] The optimal operative methods and extent of resection are also controversial,[10] but recurrence is lower in patients undergoing complete resection.[10]
We present herein a rare case of a 27-year-old Indonesian woman who had a retropharyngeal abscess with cervical Pott disease and tuberculous abscess of the chest wall without active pulmonary tuberculosis.

2. Case report

A 27-year-old woman, who was a foreign domestic worker from Indonesia, came to our outpatient department of Family Medicine in December 2015 with a chief complaint of pain in the right side of the neck for 2 weeks. The pain was intermittent, aggravated with neck rotation, and unrelieved by analgesia, and its severity had gradually increased. She had a sore throat, dysphagia, odynophagia, intermittent fever, body weight loss for 1 month, and a gradually enlarging mass over the right anterior chest wall for 4 months. There was no history of cough, dyspnea, paresthesia, or weakness to the upper and lower limbs, or head and neck trauma. Her only remarkable past history was left ureteral stricture with hydronephrosis, and she underwent laparoscopic pyeloplasty in August 2015. She did not take any drugs, had no family history of systemic diseases, and had no travel or contact history. On physical examination, her body temperature was 37.2°C; heart rate, 110 beats/min; and blood pressure, 115/71 mm Hg. The patient looked ill, with swelling and redness without overlying skin ulceration in the right side of the neck. Palpation of the right side of the neck revealed softness, tenderness, and heat. Examination of the oral cavity showed anterior displacement of the posterior pharyngeal wall. The mass over the right anterior chest wall measured approximately 5 × 4 cm in size. The lungs were clear to auscultation, and neurological examination revealed normal findings.

She was referred to the ear, nose, and throat (ENT) outpatient clinic. Flexible fiberoptic endoscopy revealed a bulging mass originating from the retropharyngeal space. Laboratory tests revealed a white blood cell count of 10.37 × 10^3/μL (neutrophils, 78.9%) and C-reactive protein level, 5.7 mg/dL. The antibody test for human immunodeficiency virus (HIV) yielded negative results. All other laboratory tests were normal.

Cervical spine radiography on anteroposterior and lateral view revealed a prevertebral soft tissue mass anterior to the C1 to C7 level, displacing and indenting the adjacent airway (white arrow); focal destruction of the C3 vertebra (black arrow) and straightening of the cervical spine; and ground glass opacity over the right upper lung field, located in the extrapulmonary space (Fig. 1A and B).

Chest radiography revealed an extrapulmonary mass over the right upper lung field, which was a similar finding on cervical spine radiography (Fig. 2) and was compatible with the patient’s right anterior chest wall mass.

A contrast-enhanced computed tomography (CT) scan of the neck showed multiloculated retropharyngeal fluid collections with enhancing walls spanning from the level of the nasopharynx down to the level of the supraclavicular fossa (Figs. 3A–C, 4A, and B, double-ended black arrow), extending to the prevertebral space and epidural space of the spinal canal at the levels of C1-C3 (Figs. 3B and 4B, black arrow), with destruction of the C3 vertebral body. These lesions caused significant narrowing of the oropharyngeal and hypopharyngeal airways. Destruction of the T9 vertebra from a left paraspinal mass measuring 3.2 × 2.9 × 1.9 cm was also found (Figs. 4C and 5, white arrow). Moreover, a large fluid collection with thick irregular walls over the right anterior chest wall measuring approximately 6 × 5.7 × 5.3 cm in

Figure 1. A, Cervical spine radiography, lateral view showed prevertebral soft tissue anterior to the C1 to C7 level (white arrow), focal destruction of the C3 vertebra (black arrow), and straightening of the cervical spine. B, Cervical spine radiography, anteroposterior view showed a mass displacing and indenting the adjacent airway (white arrow) and extrapulmonary opacity over the right upper lung field.
size with the destruction of the anterior part of the right second rib was noted, and it extended to the extrapulmonary intrathoracic cavity (Fig. 6). These findings were suggestive of retropharyngeal abscess extending to the cervical spine and chest wall abscess.

The patient was admitted to the ENT ward for further operative management, and a thoracic surgeon at our hospital was consulted for the management of the chest wall abscess. Preoperatively, she received intravenous amoxicillin/clavulanic acid (1200mg) every 8 hours and gentamycin (80mg) every 8 hours. Surgery was performed on the second day of hospitalization, and drainage of the right retropharyngeal abscess and chest wall abscess was performed simultaneously. Pus cultures yielded negative results for common bacteria. Acid-fast stain (AFS) yielded positive results. Polymerase chain reaction (PCR) yielded positive findings for Mycobacterium tuberculosis complex. Histopathology showed chronic caseating granulomatous inflammation and Langhans multinucleated giant cells. Pus cultures revealed M tuberculosis complex after 3 weeks of inoculation. Three sputum samples all tested negative for acid-fast bacilli, and cultures were negative for M tuberculosis. Based on these findings, a diagnosis of tuberculous retropharyngeal abscess with Pott disease and chest wall abscess was established.

The patient received isoniazid (300mg), rifampicin (600mg), pyrazinamide (1500mg), and ethambutol (800mg) daily. Antituberculous drug sensitivity test showed that the M tuberculosis complex was susceptible to these medications. Laboratory examination revealed a white blood cell count of $9.51 \times 10^3/\mu\text{L}$ (neutrophils, 88%) and C-reactive protein level, 4.74mg/dL. All other laboratory test results were normal on postoperative day 1. Postoperative chest radiography revealed marked regression of the chest wall abscess as compared with the preoperative image. The patient made an uneventful postoperative recovery with considerable improvement and resolution of both abscesses. She was discharged on postoperative day 13 and returned home to Indonesia.

2.1. Ethical statement and consent

The patient returned to her home country after discharge, and so we were unable to obtain her informed consent. The need for obtaining informed consent was waived off as per institutional review board of Taipei Tzu Chi Hospital, Buddhist Tzu Chi Medical Foundation. This case report was approved by the institutional review board (approval number: 07-CR-099) of Taipei Tzu Chi Hospital.

3. Discussion

The retropharyngeal space, cervical spine, and chest wall are all uncommon locations of extrapulmonary tuberculosis and are all
difficult to diagnose. The retropharyngeal space is a potential space of the neck and head. It extends from the base of the skull to the upper mediastinum and lies between the pharynx and prevertebral musculature. It is bounded by the fascia anteriorly and posteriorly and laterally by the carotid space. This space contains lymph nodes that are normally present in children only and regress during puberty. Retropharyngeal abscess can present as an acute or chronic condition. Acute retropharyngeal abscess is usually seen in children younger than 5 years old and is commonly caused by infection of the retropharyngeal lymph nodes that drain lymph from the ear, nose, and throat. Acute retropharyngeal abscess in adults is often pyogenic and may result from penetrating injury, foreign body, endotracheal intubation, or endoscopic procedures. Chronic retropharyngeal abscess is usually of tuberculous etiology and is commonly seen in adults. It is usually due to the tuberculous involvement of the cervical spine as pus erodes into the retropharyngeal space on both sides of the midline. Chronic retropharyngeal abscess can also be caused by tuberculous infection involving the retropharyngeal lymph nodes, which is usually limited to one side.
of the midline.\[2\] The possible route of spread of tuberculosis to the retropharyngeal space is via the lymphatics to a persisting retropharyngeal lymph node.\[13\] In rare cases, the abscess may be caused by hematogenous spread from pulmonary tuberculosis or tuberculosis elsewhere.\[13\] Retropharyngeal abscess caused by tuberculosis is rare, even in the presence of extensive pulmonary tuberculosis.\[14\]

Approximately 1% of all patients hospitalized with tuberculosis have skeletal tuberculosis, with only 7% of these involving the cervical spine.\[13\] When skeletal tuberculosis involves the spine, the most common site would be the lumbar vertebrae, followed by the thoracic and cervical vertebrae.\[13\] This supports the theory that infection in Pott disease begins in the pelvic organs and disseminates hematogenously via Batson plexus to involve more superior parts of the vertebrae in a watershed fashion.\[11\] Odynophagia and dysphagia are principal symptoms of retropharyngeal abscess. Our patient presented with these symptoms. Neck rigidity, external neck swelling, and airway obstruction may occur in more severe cases.\[4\] A deep neck abscess may be life-threatening due to the possibilities of involvement of the carotid sheath, airway obstruction, tracheobronchial aspiration via spontaneous rupture of abscess, spread into the mediastinum, or septic shock.\[16\] The mortality rate with these fatal complications is as high as 40% to 50%.\[16\] The classic symptoms of tuberculosis such as night sweats, body weight loss, and cachexia may be present in the early stage of Pott disease,\[17\] but they are not usually found in tuberculous retropharyngeal abscess.\[4\] Patients with Pott disease may have pain at the back of the neck and restricted neck movements. If the abscess expands, it may compress the spinal cord and cause neurological deficit, or bulge anteriorly and lead to airway obstruction.\[12\]

Early diagnosis is essential to prevent the complication of retropharyngeal abscess and Pott disease. It should be suspected in patients who present with a retropharyngeal mass and a destructive lesion of the vertebra.\[2\] However, the clinical diagnosis is difficult due to nonspecific symptoms, such as dysphagia. Dysphagia itself is common in the population, particularly among the elderly, who are also at higher risk of tuberculosis. Diagnosis is mainly based on a high index of clinical suspicion particularly in an endemic area, radiological features, aspiration of the mass for bacteriologic culture, biopsy for histopathologic examination, and PCR.\[4\] The incidence of chronic retropharyngeal abscess is increasing because of the resurgence of tuberculosis secondary to HIV infection; thus, all patients should be screened for this disease.\[11\] Moreover, the use of immunosuppressive medication is also one of the causative factors, particularly in developed countries. Lateral view radiography of the neck can be helpful in the diagnosis. Individuals with a widening of or air in the prevertebral soft tissues, osteolytic lesion in the cervical vertebrae, or loss of the cervical spine lordosis or vertebral height should be suspected of the condition.\[1,4\] The normal depth of the retropharyngeal soft tissue overlying the C2 averages 4 mm, and soft tissue greater than 7 mm is pathologic.\[16\] Chest radiograph is useful in screening pulmonary tuberculosis or complications such as mediastinitis, pneumonia, and pleural effusion. Ultrasoundography is an affordable, simple, and nonirradiating choice of examination, but a negative result cannot rule out retropharyngeal abscess.\[4\] Meanwhile, CT scan is a useful diagnostic tool that can confirm the abscess location, size, extensions, and Pott disease.\[4\] A contrast-enhanced CT scan finding of a central hypodensity mass with surrounding ring enhancement suggests the presence of an abscess.\[16\] Lateral view radiography of the neck has sensitivity and specificity of 80% and 100% in diagnosing retropharyngeal abscess, while for a CT scan, it is 100% and 45%, respectively.\[2\] However, CT scan has a limitation in differentiating cellulitis from abscess with accuracy of 73.5%, false-positive rate of 11.8%, and false-negative rate of 14.7%.\[2\] The decision for drainage or surgical intervention must be guided by CT findings and diagnosis.\[2\] Meanwhile, magnetic resonance imaging (MRI) findings of low signal intensity on T1-weighted images and high signal intensity on T2-weighted images also suggest an abscess,\[16\] and MRI can diagnose any complications more precisely, such as vein thrombosis.\[4\]

Early treatment includes antitubercular medication and surgical intervention, and the combination of these treatments may be optimal. If an abscess is large enough to be detected on clinical examination, drainage should be performed.\[16\] Surgical drainage includes the simple intraoral approach, extensive external cervical approach, or minimally invasive technique such as image-guided needle aspiration.\[17\] The need for external drainage for retropharyngeal abscess has been controversial.\[16\] Needle aspiration has several advantages. It can be repeated if needed, can be performed under local anesthesia, and can be used to rule out other differential diagnoses such as a malignant tumor presenting clinicoradiologically as an abscess.\[18\] In patients whose pus cannot be successfully and completely aspirated, external drainage may be performed. Antituberculous medication and conservative neck stabilization should be the initial treatment for Pott disease.\[19\] Surgical intervention in Pott disease is indicated if there is a neurologic deficit, spinal deformity with instability or pain, no response to antitubercular chemotherapy, and large paraspinal abscess.\[20\] Majority of patients with tuberculous retropharyngeal abscesses with neurologic complications recover following prompt surgical drainage and antitubercular medications.\[21\]

Skeletal tuberculosis is rare, accounting for only 2.6% of all tuberculosis cases.\[15\] Most cases of skeletal tuberculosis occur in the vertebrae, hips, and knees.\[8\] Tuberculous involvement of the chest wall is uncommon and accounts for <10% of cases of skeletal tuberculosis.\[13\] Abscesses of the chest wall are usually solitary, but multiple lesions can also be present.\[5\] Abscesses are more frequently found in the shaft of the ribs and at the parasternal area.\[5\] The costovertebral junction and vertebrae are involved less frequently. Tuberculous involvement of the chest wall has been reported to occur more frequently in men (male/female ratio: 1:2.1−8:1).\[17,54\] The commonly affected age group varies in studies.\[9\] Some authors reported a more frequent incidence at a younger age (15−35 years), whereas others reported a higher rate at an older age.\[13\] In Faure’s study, 83% of cases had a past history of tuberculosis, and the incidence of active pulmonary tuberculosis ranged from 17.4% to 62.5%. Three mechanisms underlie the pathogenesis of rib tuberculosis: direct invasion from adjacent pleural or pulmonary parenchymal tuberculosis, hematogenous dissemination from activation of a dormant tuberculous focus, and local extension from lymphadenitis of the chest wall.

The first mechanism is considered to be the least common mechanism, whereas the second is the most common.\[6,7\] Skeletal tuberculosis is believed to result from either hematogenous or lymphatic dissemination of bacilli from the site of the primary infection in the lung.\[22\]

The clinical presentation of abscess of the chest wall is a progressively enlarging and sometimes painful mass with or
without destruction of the underlying bone or cartilage and with or without signs of local inflammation.[8] The consistency of the mass may be firm or soft. Differentiating a tuberculous abscess of the chest wall from a pyogenic abscess or chest wall tumor may be difficult.[8] Diagnosis may be challenging due to the absence of specific signs and symptoms and requires a high index of clinical suspicion. Needle aspiration may be the procedure of choice as it is less invasive to establish the diagnosis of tuberculosis and to rule out other conditions such as malignancy or other infection.[5] It may show acid-fast bacilli in the direct smear or in the culture. However, this examination can only establish a definite diagnosis in approximately 20% of patients.[10] Performing PCR with a smear or culture has a higher sensitivity.[10] Ultrasonography is advantageous in that it is radiation free, and it can demonstrate infection focus and be used for ultrasound-guided needle biopsy at the bedside. Chest radiography may show osteolytic lesions with overlying dense opacities, but it may not be consistent with tuberculosis and rather reveal the possibilities of malignancies, either primary or metastatic.[23] CT is a useful tool in the diagnosis of abscess of the chest wall, in defining the extent of the abscess, and in confirming the appropriate site for biopsy. Common chest CT findings are juxtaocular soft tissue mass with central low attenuation, peripheral rim enhancement, calcification, and bone or cartilage destruction.[9] However, these CT findings are also nonspecific. MRI is helpful in differentiating between abscess and granulation tissue, but it is not the first examination modality of choice.[24] Radionuclide bone scan is also nonspecific because it cannot differentiate between malignancy and tuberculous lesions, and findings may be negative in the early stages of the disease.[24] Laboratory examinations, such as complete blood count, erythrocyte sedimentation rate, and C-reactive protein are usually unremarkable.

Treatment should be started immediately after the microbiological and histological samples have been obtained if the clinical suspicion is high, particularly when there is epidemiologic data on tuberculosis infection. However, diagnosis must still be confirmed via culture or histology. The optimal treatment of abscess of the chest wall is controversial.[5] Most authors recommend combining medical and surgical management to increase efficacy and to prevent recurrence.[8] The abscess is rarely treated using antituberculous medication only,[9] and it is difficult to decide whether a patient can be successfully treated without operation. The size of the abscess, CT findings, or comorbidities do not appear to correlate entirely with the clinical course.[10] The proper methods of operative treatments and extent of resection are also controversial.[10] The optimal approach consists in excising the abscess and performing a primary closure; the abscess should not be left open as that favors chronic sinus formation.[5] Kaufmann suggested resecting a part of the rib even if it is macroscopically normal because the origin of the abscess is deeper[22,23] and because more than half of the lesions of rib tuberculosis show no evidence of rib destruction.[24] Therefore, Brown and Trenton[27] also suggested extensive debridement of the lesion, particularly if there is involvement of the cartilage and bone. However, Weissberg[28] preferred drainage of the abscess for the tissue diagnosis followed by antituberculous medication, reserving debridement and excision only for the most extensive diseases. Intraoperative finding and evaluation of the extent of the lesion is important because preoperative imaging does not always reveal the involvement of the pleura and ribs.[10] Most authors recommend antituberculous medication after surgical resection. However, the completeness of the resection of the abscess is more important in determining the development of recurrence.[9] In Paik et al’s[8] study, the recurrence rate was 16% after excision only of the abscess compared with the 4.7% after complete resection of the mass including any involved ribs. In Kim et al’s[9] study, the recurrence rate was higher in the drainage only group compared with that in the complete resection group (40.0% vs 9.2%). The World Health Organization recommends a standard 6-month regimen if the patient received complete surgical resection.[22,29] However, according to clinical presentation, bacillary load, response to antituberculous medication, and whether surgery was performed or not, the treatment duration can be extended up to 9 to 12 months.[22,29] Isoniazid (H), ethambutol (E), rifampin (R), and pyrazinamide (Z) for 2 months followed by at least 4 months of HER is the most commonly used regimen.[8] Recurrence may be due to insufficient resection, new lesion, drug-resistant strain, or nonadherence to medication.[8] Tuberculosis is the great mimicker; it presents differently and affects many organ systems.[7] Its diagnosis remains challenging due to the lack of specificity of symptoms and requires a high index of clinical suspicion with a proper history and physical examination. Laboratory investigations (AFS, PCR, culture), radiography (x-ray, CT scan), and ultrasonography are useful diagnostic tools. Our patient presented with nonspecific symptoms. We were aware of the possibility of tuberculosis because she came from Indonesia where the incidence and prevalence were relatively higher than those of developed countries. Her retropharyngeal mass and destructive lesion of the vertebra were also suggestive of a tuberculous etiology.[31] Drainage of the retropharyngeal abscess was indicated because her abscess was large enough to cause symptoms and airway compression.[16] Incision and drainage were chosen rather than needle aspiration because drainage with debridement of the chest wall abscess could be performed under general anesthesia simultaneously. Postoperatively, she received antituberculous drugs and the PCR yielded positive results for M tuberculosis complex. Postoperatively, her symptoms improved, and both abscesses disappeared without wound infection or bleeding during hospitalization. However, she returned to her home country after discharge. We could not follow-up in more detail about her adherence to medication, long-term treatment outcomes, and possible complications or recurrence. The retropharyngeal space, cervical spine, and chest wall are all uncommon locations of extrapulmonary tuberculosis.[5,14,15] This case is unique in that the patient had extrapulmonary tuberculosis involving 3 rare sites simultaneously. Tuberculosis is a considerable health burden globally. Early diagnosis and treatment are necessary to prevent life-threatening complications. Physicians should keep tuberculosis in mind when patients present similarly. Most clinicians recommend a combination of antituberculous medication and surgical intervention.[8] However, there are no guidelines regarding the operative management of tuberculosis retropharyngeal and chest wall abscess.[9] The extent of surgical resection of chest wall abscess remains controversial,[10] and the recurrence rate is lower in the complete resection group as proven by studies.[8,9] Physicians should determine the optimal surgical options on a case-by-case basis, according to age, comorbidity, extent of the lesion, and intraoperative findings[10], etc. Nonadherence to antituberculous drugs is one of the reasons for recurrence; thus, clinicians should educate patients and follow them up to monitor treatment outcomes, possible complications, or recurrence.
4. Conclusions

Tuberculous retropharyngeal abscess is a rare form of extrapulmonary tuberculosis. A possibility of tuberculous etiology should always be considered in patients presenting with abscess regardless of the presence or absence of clinical and radiological features suggestive of tuberculosis. Proper history and examination are important for early diagnosis. Aggressive treatment with a combination of antituberculous medication and early surgical intervention are necessary to prevent life-threatening complications.

Tuberculous abscess of the chest wall is uncommon, and its diagnosis remains challenging and requires a high index of clinical suspicion. The most appropriate diagnostic and management plan may be performing a needle aspiration or biopsy first. If a tuberculous infection is strongly suspected or confirmed, a combination regimen of antituberculous medication and surgical resection should be started. Complete resection and postoperative antituberculous medication for a minimum of 6 months are important to prevent recurrence.

Author contributions

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