



A rare case of chest wall tuberculosis: Tuberculous scapulothoracic bursitis

Norichika Iga^{a,*}, Yasuko Fuchimoto^b, Taisaku Koyanagi^b, Daisuke Mizuno^a, Hideyuki Nishi^a

^a Department of Surgery, Okayama Rosai Hospital, 1-10-25 Chikkomidorimachi, Minamiku, Okayama, 702-8055, Japan

^b Department of Respiratory medicine, Okayama Rosai Hospital, 1-10-25 Chikkomidorimachi, Minamiku, Okayama, 702-8055, Japan

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ABSTRACT

Chest wall tuberculosis is a relatively rare extrapulmonary tuberculosis, and is often difficult to diagnose and treat because of the lack of symptoms. The scapulothoracic joint is a special joint that does not have a joint capsule, cartilage, or synovial membrane but consists of muscle and bursa. Tuberculosis infection of the scapulothoracic joint is an extremely rare musculoskeletal tuberculosis of the chest wall. Herein, we present the diagnosis and treatment strategy for tuberculous scapulothoracic bursitis in an 82-year-old man who was successfully treated.

1. Introduction

Chest wall tuberculosis is an extrapulmonary tuberculosis, accounting for approximately 1%–2% of all cases of tuberculosis [1]. Chest wall tuberculosis often presents with cold abscesses, has no specific features or symptoms, is difficult to diagnose and treat, and is often misdiagnosed as another benign or malignant disease of the thorax [2]. Antituberculous chemotherapy alone is not effective in the treatment of chest wall tuberculosis, and a combination of chemotherapy and surgical resection is recommended [2,3]. However, chest wall abscesses occasionally lead to cutaneous fistulas and destruction of bone and cartilage, and the surgical approach and extent of resection require careful planning.

Unlike other shoulder joints, the scapulothoracic joint is not covered by a joint capsule, cartilage, or synovial membrane. It is composed of muscles and bursae located between the scapula and chest wall, allowing smooth gliding motion. Scapulothoracic bursitis is a rare disorder caused by inflammation of the bursa secondary to trauma or overuse due to sports activities or work [4]. Herein, we report a case of chest wall tuberculosis involving the scapulothoracic joint. Tuberculous scapulothoracic bursitis has never been reported and is extremely rare, and we present the steps in the diagnosis and treatment of the same.

2. Case report

2.1. Patient information and clinical findings

An 82-year-old man was referred to our hospital with a complaint of a bulge in the left chest wall. On palpation, the cystic mass was soft and

highly flexible, without redness or heat. The patient had a history of left pleural effusion developed one year prior, and thoracoscopic examination had been performed because the adenosine deaminase (ADA) level in the pleural effusion was high (97.4 U/L). Malignant pleural diseases and tuberculous pleuritis had been ruled out by bacteriological and histological examination. Six months after thoracoscopy, computed tomography showed that the pleural effusion had decreased in volume, but there were numerous small nodular shadows in the left upper lobe; the nodules had increased in size and showed a tree in bud appearance, and a cystic mass appeared under the left scapula (Fig. 1). The patient had undergone surgery for lumbar spinal canal stenosis 13 years earlier, and needed a walking frame because of the complications of post-operative leg paresis. In addition, there was an episode of rehabilitation of shoulder movement performed three times a week for one year after the last thoracoscopic examination; treatment was discontinued due to shoulder pain.

2.2. Diagnostic assessment

Ultrasonography showed that the cystic mass had a wall of approximately 5 mm and contained numerous hyperechoic deposits (Fig. 2). Magnetic resonance imaging showed that the cystic mass was 12 cm in size and located inside the serratus anterior muscle below the scapula, and there were many crystalline structures inside the cystic mass that showed low signal intensity on T2-weighted images, suggesting scapulothoracic bursitis (Fig. 2). The white blood cell count and C-reactive protein levels were 5800/ μ l and 0.2 mg/dL, respectively. The erythrocyte sedimentation rate was 13 mm at 1 h, and T-SPOT. TB test was

* Corresponding author.

E-mail address: iga-toku@okayamah.johas.go.jp (N. Iga).

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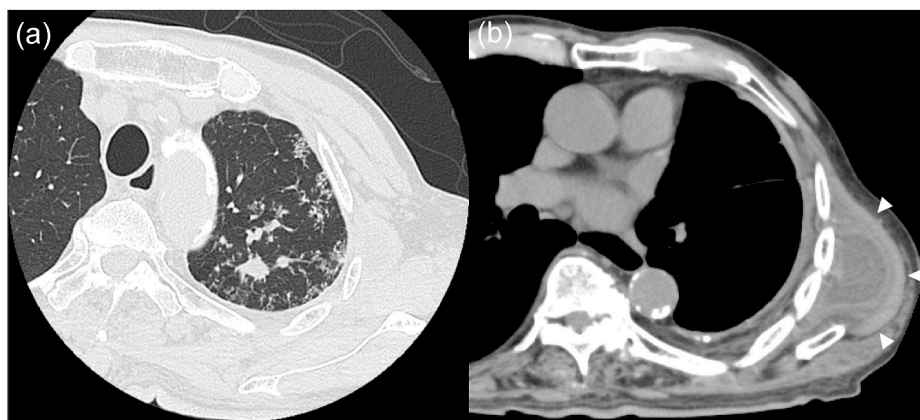


Fig. 1. (a): Chest computed tomography showing a small nodular shadow with a tree-in-bud appearance in the left upper lobe. (b): Chest computed tomography showing encapsulated fluid in the chest wall.

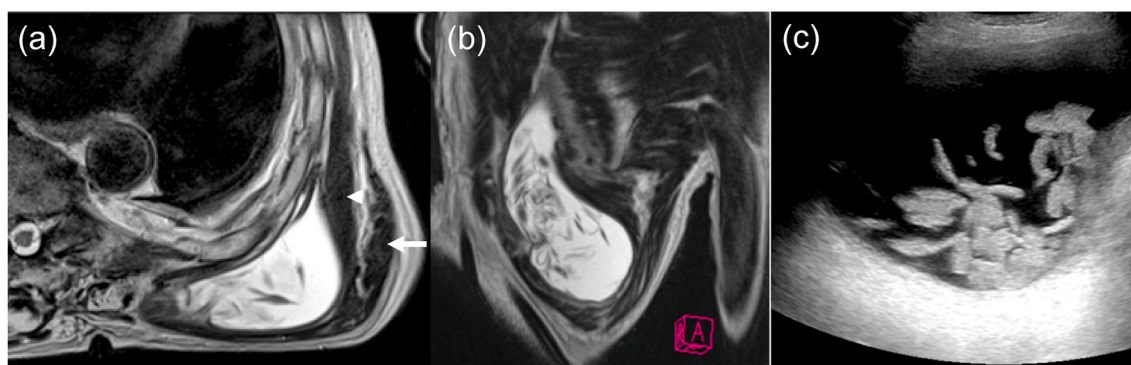


Fig. 2. Magnetic resonance imaging and Ultrasonography imaging of chest wall: (a): An axial T2-weighted image showing a cystic mass with high-signal-intensity fluid deep in the serratus anterior muscle. Arrow head: serratus anterior muscle. Arrow: Latissimus dorsi muscle. (b): A coronal T2-weighted image showing numerous low-signal-intensity trabecular structures within the cystic mass. (c): Cystic mass contains hyperechoic deposits.

Table 1
Biochemical findings of cyst contents.

Appearance	orange color	Total protein	4.6	g/dL
pH	7.257	LDH	1077	IU/L
Specific gravity	1.033	Gulcose	5	mg/dL
Cell classification		ADA	99.1	U/L
Nentrophil	25%	CEA	3.3	ng/mL
Macrophage	3.00%	CA19-9	5.7	U/mL
Lymphocyte	72.00%	CYFRA	0.5	ng/mL

LDH, Lactate dehydrogenase; ADA, Adenosine Deaminase. CEA, Carcinoembryonic antigen; CA19-9, Carbohydrate antigen19-9. CYFRA, Cytokeratin fragment.

negative. Biochemical and microbiological examination of the cyst contents by needle aspiration and histological examination of the cyst wall by needle biopsy were performed to diagnose the cystic mass. The fluid in the cyst was orange and slightly turbid, and the ADA level in the cyst fluid was elevated (99.1). The results of the biochemical examination of the fluid contents are presented in [Table 1](#). *Mycobacterium tuberculosis* was detected by a PCR test from the contents, and multinucleated giant cell infiltration was observed in the cyst wall.

2.3. Therapeutic management and surgical procedure

Since there were no malignant findings, the patient was diagnosed with tuberculous scapulothoracic bursitis, and antituberculous therapy

(rifampicin 450 mg, isoniazid 300 mg, ethambutol 750 mg) was initially started. However, imaging evaluation after 3 months of antituberculosis drug treatment showed enlargement of the cystic mass. Therefore, we decided to perform surgical excision.

The operative team donned N 95 masks, considering the rupture of the cystic mass. Under general anesthesia in the right lower decubitus position, a skin incision was made along the anterior border of the latissimus dorsi muscle from the axilla to the caudal margin of the mass. The serratus anterior muscle was split along the muscle bundle at the caudal side of the mass to reach the mass. In most places, the cystic mass was loosely adhered to the surrounding tissue, but part of the cranio-dorsal region was adhered to the fourth intercostal muscles, so the surface layer of the periosteum and intercostal muscle was resected. The cystic mass was resected without rupture (Video 1).

Supplementary video related to this article can be found at <https://doi.org/10.1016/j.rmcr.2021.101537>

2.4. Specimen and histological findings

The cystic mass was composed of a uniformly fibrous wall containing numerous coin-shaped chondroid bodies ([Fig. 3](#)). Histological findings showed inflammatory cell infiltration, including lymphocytes, histiocytes, and plasma cells, inside the fibrous cyst wall. A granulomatous lesion with clusters of multinucleated giant cells, which was not accompanied by necrosis but suggested tuberculosis infection, was observed ([Fig. 4](#)).

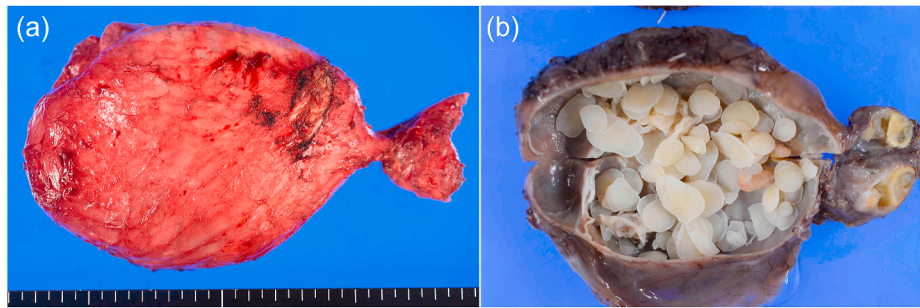


Fig. 3. Macroscopic findings of the resected specimen:

The dumbbell-shaped cystic mass, measuring 17 × 10 cm, contained serous fluid and numerous coin-shaped fibrin deposits were observed inside.

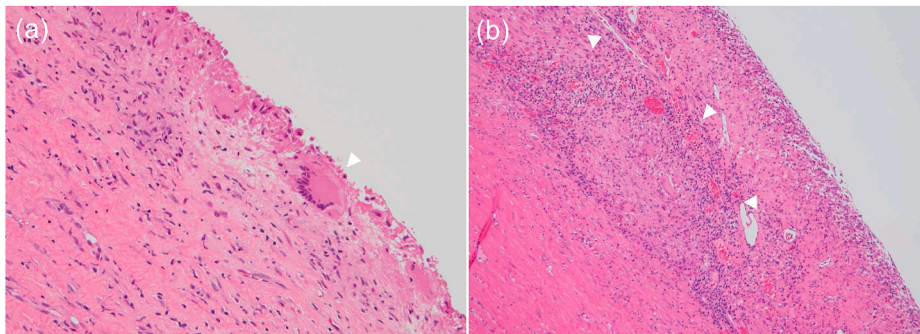


Fig. 4. Microscopic findings of the cyst wall:

(a): the cyst wall obtained by needle biopsy before antituberculous therapy. Multinucleated giant cells (arrow head) infiltrate the fibrous cyst wall.

(b): Cyst wall of the resected specimen after antituberculous therapy

The cyst wall is infiltrated with inflammatory cells such as lymphocytes, histiocytes, and plasma cells. Granulomatous lesions (arrowheads) with aggregated multinucleated giant cells are observed.

2.5. Follow up

As postoperative adjuvant therapy, the three antituberculosis drugs mentioned in section 2.3 were administered for 6 months. The chest wall cyst did not recur, the small nodular shadow in the left upper lobe improved, and exacerbation of tuberculosis was not observed for more than one year postoperatively.

3. Discussion

Chest wall tuberculosis is an extrapulmonary tuberculosis that accounts for 1%–5% of all cases with musculoskeletal tuberculosis, representing 1%–2% of all cases of tuberculosis [1].

Many patients with chest wall tuberculosis have a history of treatment for tuberculosis or concomitant tuberculosis [3]. Common sites of skeletal tuberculosis are the spine, hip joint, knee joint, foot, elbow, hand, and shoulders [1].

Unlike other joints, the scapulothoracic joint between the scapula and thorax, does not have a joint capsule, cartilage, or synovial membrane, but is composed of muscle and bursa, allowing smooth, gliding motion [4]. Two major and four minor bursae at the scapulothoracic articulation have been described [5]. Scapulothoracic bursitis is caused by inflammation secondary to trauma or repeated mechanical overuse, such as sports or work [6]. Multiple cartilaginous bodies (rice bodies) are observed in various joints and bursae. They are termed as rice bodies because of their close resemblance to the shape and size of the polished rice grains. Rice bodies were first described by Riese in association with tuberculosis arthritis in 1895 [7]. Among the diseases in which the rice bodies are observed in the joint, rheumatoid arthritis is the most common, but nonspecific inflammation, and infectious arthritis, such as tuberculosis have also been reported. Albrecht et al. [8] postulated that the rice bodies are the end product of synovial inflammation, proliferation, and subsequent secondary degeneration leading to sloughing of the synovial membrane, which becomes covered with fibrin derived from the synovial fluid. The characteristics of rice bodies on magnetic resonance imaging are useful for diagnostic clues because they are

depicted as intermediate signals on T1-weighted images and low-signal nodes on T2-weighted images [9].

In the current case, scapulothoracic bursitis was caused by tuberculosis infection. Tuberculous infection of the scapulothoracic joint has never been reported and is extremely rare. Unlike a tuberculous chest wall abscess, the abscess in the scapulothoracic bursa was movable, had very loose adhesions with surrounding tissues, and was easily excised completely. Three mechanisms have been described to explain the pathogenesis of chest wall tuberculosis: direct extension of pulmonary or pleural involvement, hematogenous dissemination, direct transcutaneous inoculation, or extension from lymphadenitis of the chest wall [3]. The lesion in this case was considered to have been caused by direct extension of the lung or pleural lesion. A left pleural effusion was examined one year prior, and no evidence of *Mycobacterium tuberculosis* was found from the effusion or biopsied pleura, but it is presumed that there was latent tuberculous pleuritis at that time, and direct pleural spread of tuberculosis to the scapulothoracic bursa was caused by overuse of the shoulder in the same period. Tuberculosis PCR testing performed during thoracoscopy was performed with the Cobas TaqMan MTB (Roche Diagnostics). The sensitivity and specificity of Cobas TaqMan MTB PCR have been found to be 79.1% and 98.2%, respectively [10]. The sensitivity and specificity of Xpert MTB/RIF (Beckman Coulter) are 51.4% and 98.6%, respectively, based on the culture method, and 22.7% and 99.8%, respectively, based on a composite reference standard [11]. Although the sensitivity is low, the specificity is high, and it might have been possible to make the diagnosis using this reagent. In this case, the pleural fluid ADA level was high during thoracoscopy. Aggarwal et al. [12] reported that the sensitivity and specificity of ADA in pleural fluid for the diagnosis of tuberculous pleural effusion were 86% and 94%, respectively, when the level of ADA in pleural fluid was 65 U/L or more. This case was diagnosed as tuberculous pleural effusion due to the high level of ADA (97.4 U/L) in the pleural effusion, and if antituberculosis treatment had been started, the development of chest wall tuberculosis, as seen in this case, might have been prevented.

With regard to treatment of chest wall tuberculosis, Cho et al. [3]

reported that antituberculosis drugs alone are not recommended, and many authors recommend a combination of medical and surgical management. In addition, Cho reported that relapse was more common in patients who underwent surgery without preoperative antituberculosis therapy, and 6–12 months of postoperative antituberculosis therapy was administered to all patients who underwent surgery.

Faure et al. [13] also reported that if tuberculosis infection is confirmed or strongly suspected, a combination of antituberculous chemotherapy should be considered as the initial treatment and that surgical intervention is indicated if the lesion does not improve or worsens after 1–3 months of therapy. In the present case, antituberculous therapy was administered for 4 months preoperatively, and 6 months postoperatively, with reference to the above literature.

4. Conclusion

This report describes a case of tuberculous scapulothoracic bursitis. Surgical excision with antituberculous therapy proved to be an effective treatment strategy. No recurrence was observed for 22 months after the start of treatment, and a good course was obtained.

Consent for publication

Informed consent was obtained from the patient for the publication of this case report.

Competing interests

The authors declare that they have no competing interests.

Declaration of competing interest

The authors declare no conflicts of interest associated with this

manuscript.

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