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#### CASE REPORT

# Rare case of cystic anterior mediastinal tuberculosis in an immunocompetent patient

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#### Abstract

We report a case of a 28-year-old immunocompetent woman found to have a mediastinal lesion on chest x-ray performed as part of a visa renewal process. Computed tomographic imaging revealed a cystic superior anterior mediastinal mass. Although initially asymptomatic, the woman subsequently developed progressive chest discomfort. She underwent surgical resection of the mass. Histological assessment demonstrated necrotizing granulomatous inflammation, while Gene Xpert<sup>TM</sup> testing was positive for *Mycobacterium tuberculosis* complex and she was subsequently commenced on anti-tuberculous therapy.

KEYWORDS cystic, mass, mediastinal, *Mycobacterium tuberculosis* 

## INTRODUCTION

In 2018, 87% of new tuberculosis (TB) cases in Australia occurred in individuals born overseas.<sup>1</sup> Thirty-six percent were diagnosed with extrapulmonary TB, with half of these individuals having disease limited to lymph nodes.<sup>1</sup> Isolated mediastinal tuberculous lymphadenopathy is predominantly seen in the paediatric cohort and is rare in adults.<sup>2</sup>

The differential diagnosis for a mediastinal mass is broad as many structures pass through this anatomical area including the thymus, lymph nodes, adipose tissue, nerves, vessels and occasionally the thyroid gland.<sup>3</sup> The most common causes for a mediastinal mass are thymoma, teratoma, thyroid disease, lymphoma and lymph node enlargement secondary to metastatic or sarcoidosis (noting that this is normally symmetrical and bilateral lymph nodes).<sup>3</sup> The use of computed tomography (CT) can differentiate potential causes based on the predominant attenuation values present (e.g., increased presence of fat with a lipoma or teratoma). However, biopsy and/or surgical excision remains the gold standard diagnostic modality.

In particular, the causes of cystic mediastinal masses (higher water attenuation values on imaging) include congenital benign cysts, meningocele, mature cystic teratoma and lymphangioma. Other causes, including tumours, abscesses and pseudocysts, can degenerate and cause a partially cystic appearance.  $^{4,5}$ 

Many patients remain asymptomatic until they develop symptoms caused by compression of adjacent structures and are thus incidentally diagnosed.

#### **CASE REPORT**

A 28-year-old asymptomatic woman with no past medical history was referred with abnormal findings on a chest x-ray (CXR; Figure 1A) performed as part of a visa renewal process. Incidentally, a CXR performed on her arrival to Australia from New Delhi, India, 2 years prior had not revealed any abnormalities. A CT chest was subsequently organized and revealed a right paratracheal superior mediastinal mass measuring  $65 \times 64 \times 73$  mm (Figure 1B). Her immunoglobulin release assay was positive (TB antigen 1 0.54, TB antigen 2 0.76, mitogen index 6.53). She was referred for surgical excision; however, her surgery was deferred as she was asymptomatic.

The patient represented 2 months later with right-sided chest pain but no associated fevers, night sweats and weight loss. Notably, there were no symptoms to suggest superior vena cava (SVC) obstruction or airway obstruction. Full

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**FIGURE 1** (A) Chest x-ray—right-sided mediastinal mass. (B) Computed tomography chest—right paratracheal superior mediastinal, well-circumscribed lesion measures approximately  $64 \times 69 \times 75$  mm and is predominantly cystic in nature. (C) Magnetic resonance imaging (MRI) chest—T1-weighted image, predominantly cystic right paratracheal lesion with a thick enhancing wall at the superomedial margin and enhancing internal septations. (D) MRI chest—T2-weighted image. (E) Positron emission tomography —marked fluorodeoxyglucose avidity in the periphery of the large right-sided mediastinal mass. (F) Haematoxylin and eosin stain of tissue of mediastinal mass demonstrating granuloma with central caseation. (G) Macroscopic appearance of well-circumscribed, encapsulated mediastinal mass

blood count showed a white cell count of 9.66, lymphocyte count of 2.29 and C-reactive protein of 13.9.

Magnetic resonance imaging (MRI) of the chest was performed, which demonstrated a predominantly cystic lesion with thick wall enhancement at the superomedial margin and enhancing internal septations (Figure 1C—T1-weighted image, Figure 1D—T2-weighted image). Whole-body positron emission tomography scan demonstrated marked fluorodeoxyglucose (FDG) avidity in the periphery of the lesion, with a standardized uptake value max up to 17.7. The central component of the lesion was devoid of FDG uptake and there were no distant foci of FDG avidity (Figure 1E).

Praziquantel and albendazole were commenced 2 weeks pre-operatively to provide coverage for cystic echinococcosis. This was recommended to decrease the burden and risk of seeding intra-operatively despite negative serology for echinococcosis. The patient underwent a right video-assisted thoracoscopic surgery, thoracotomy and complete excision of the right paratracheal superior mediastinal mass in addition to the station 2R and 4R lymph nodes. Post-operatively, the patient developed a chylothorax that resolved with a minimal long-chain fat diet for 5 days.

The histological diagnosis of necrotizing granulomatous inflammation was highly suggestive of mycobacterial disease with lymph node tissue at the periphery and large expanses of necrosis which appeared caseating (Figure 1F). The 2R node demonstrated granulomatous lymphadenitis without necrosis and the 4R node showed a benign lymph node with no evidence of granulomatous inflammation or malignancy. There was no cystic component seen histologically. The

gross appearance was of a well-circumscribed mass (Figure 1G). Gene Xpert<sup>TM</sup> was performed on the tissue and was positive for *Mycobacterium tuberculosis* complex without the detection of RPO-B mutation conferring likely rifampicin sensitivity. *Mycobacterium tuberculosis* was cultured at 2 weeks, with drug sensitivity profiling showing sensitivity to standard anti-tuberculous treatment. She was commenced on a regimen of isoniazid, rifampicin, ethambutol, pyrazinamide and pyridoxine for 2 months, followed by isoniazid, rifampicin and pyridoxine for a further 4 months.

#### DISCUSSION

Pulmonary TB presenting as an isolated mass in the anterior mediastinum in the absence of other pulmonary lesions is rare, particularly in adults.

The initial differential diagnoses for the mass were broad with TB and hydatid cyst considered given her recent migration from India. Her main risk factor was recent migration from India where the annual incidence of cystic echinococcosis ranges from 1 to 200 per 10,000 people.<sup>6</sup> Malignant processes, such a germ cell tumour, thymoma and lymphoma, were considered less likely given the initial asymptomatic presentation and blood tests. Notably, flow cytometry was not performed due to the presence of necrotizing granulomatous inflammation on histology.

On review of the literature, seven other case reports have described cases of immunocompetent individuals, with TB presenting as an isolated mediastinal mass.<sup>4,7–12</sup> There was considerable variability in symptoms at diagnosis. Some patients were asymptomatic while others presented with local (cough, shortness of breath, chest pain) or constitutional symptoms (fever, weight loss, night sweats). One case reported dysphagia as the presenting complaint, with subsequent endoscopy demonstrating external compression of the mid oesophagus from mediastinal TB.<sup>8</sup> In our case, this patient developed right-sided chest pain which was aching in nature without associated fever, night sweats or weight loss. Notably, only two other cases reported cystic components to the mediastinal mass, emphasizing the variability in radiological appearance of mediastinal TB.<sup>8,10</sup>

The development of an isolated mediastinal mass is proposed to have developed following lymph-haematogenous dissemination of *M. tuberculosis* and due to the immunocompetent state of the patient was contained to this lymph node.<sup>13</sup> Mediastinal lymphadenopathy in the absence of pulmonary involvement can occur with TB, and the right paratracheal node is the most common location for this to occur.<sup>14</sup>

The suspected mechanism of chylothorax development in this patient was disruption to the thoracic duct as a complication of surgical instrumentation in the mediastinum.<sup>14</sup> Chyle accumulation in the pleural space was managed with the preferred methods of intercostal drain insertion and a low-fat medium-chain diet.<sup>15,16</sup>

This case highlights the need for a low threshold of clinical suspicion for TB when assessing an undifferentiated mediastinal mass, particularly in individuals from endemic regions. It must be noted that there is substantial heterogeneity in the mode of clinical presentation and radiological features of mediastinal TB.

#### AUTHOR CONTRIBUTION

**Jessica Butler:** Writing – review and editing. **Simone Barry:** Writing – review and editing.

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### **CONFLICT OF INTEREST**

None declared.

#### DATA AVAILABILITY STATEMENT

The data that support the findings of this study are available from the corresponding author upon reasonable request.

#### ETHICS STATEMENT

The authors declare that appropriate written informed consent was obtained for the publication of this manuscript and accompanying images.

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