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™ 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.1 Thirty-six percent were diagnosed with extrapulmonary TB, with half of these individuals having disease limited to lymph nodes.1 Isolated mediastinal tuberculous lymphadenopathy is predominantly seen in the paediatric cohort and is rare in adults.2

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.3 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).3 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 × 64 × 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
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 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™ 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.6 Malignant processes, such as 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.4,7–12 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.8 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.8,10

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.13 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.14

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

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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REFERENCES
1. Bright A, Denholm JT, Coulter G, Waring J, Stapledon R, National Tuberculosis Advisory Committee, for the Communicable Diseases Network Australia and the Australian Mycobacterium Reference Laboratory Network. Tuberculosis notifications in Australia, 2015–2018. Commun Dis Intell. 2020;44(1):1–27. https://doi.org/10.3321/cdir.2020.44.08
2. Agrons GA, Markowitz RI, Kramer SS. Primary tuberculosis in children. Semin Roentgenol. 1993;28:158–72.
3. Shahraz M, Le TS, Silva M, Bankier AA, Eisenberg RL. Anterior mediastinal masses. AJR Am J Roentgenol. 2014 Aug;203(2):W128–38. https://doi.org/10.2214/AJR.13.11998
4. Khilnani GC, Jain N, Hadda V, Arava SK. Anterior mediastinal mass: a rare presentation of tuberculosis. J Trop Med. 2011;2011:635385.
5. Jeung MY, Gasser B, Gangi A, Bogorin A, Charneau D, Wihlm JM, et al. Imaging of cystic masses of the mediastinum. Radiographics. 2002 Oct;22:579–93. https://doi.org/10.1148/rg.2222sup1_502009ct9
6. Akhter J, Khanam N, Rao S. Clinico epidemiological profile of hydatid disease in central India, a retrospective and prospective study. Int J Biol Med Res. 2011;2:603–6.
7. Maguire S, Chotirmall SH, Parihar V, Cormican L, Ryan C, O’Keane C, et al. Isolated anterior mediastinal tuberculosis in an immunocompetent patient. BMC Pulm Med. 2016;16:24. https://doi.org/10.1186/s12890-016-0175-7
8. Sahin F, Yıldız P. Mediastinal tuberculous lymphadenitis presenting as a mediastinal mass with dysphagia: a case report. Iran J Radiol. 2011;8(2):107–11.
9. Vincken W, Vandenbrande P, Roels P, Pirngadi J, Welch W. Isolated paratracheal mass of tuberculous origin in an adult patient. Eur J Respir Dis. 1983 Nov;64(8):630–5.
10. Jain N, Khilnani G, Hadda V, Iyer V. Cystic mediastinal mass: a rare presentation of tuberculosis. Chest. 2013;144:212A. https://doi.org/10.1378/chest.1704236

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11. Kumar N, Gera C, Philip N. Isolated mediastinal tuberculosis: a rare entity. J Assoc Physicians India. 2013 Mar;61(3):202–3.
12. Maeder M, Ammann P, Rickli H, Schoch OD. Fever and night sweats in a 22-year-old man with a mediastinal mass involving the heart. Chest. 2003 Nov;124(5):2006–9. https://doi.org/10.1378/chest.124.5.2006
13. Ganchua SKC, White AG, Klein EC, Flynn JL. Lymph nodes – the neglected battlefield in tuberculosis. PLoS Pathog. 2020;16(8):e1008632. https://doi.org/10.1371/journal.ppat.1008632
14. Mukund A, Khurana R, Bhalla AS, Gupta AK, Kabra SK. CT patterns of nodal disease in pediatric chest tuberculosis. World J Radiol. 2011;3(1):17–23. https://doi.org/10.4329/wjr.v3.i1.17
15. Nair SK, Petko M, Hayward MP. Aetiology and management of chylothorax in adults. Eur J Cardiothorac Surg. 2007 Aug;32(2):362–9.
16. Fernández Alvarez JR, Kalache KD, Grauel EL. Management of spontaneous congenital chylothorax: oral medium-chain triglycerides versus total parenteral nutrition. Am J Perinatol. 1999;16(8):415–20. https://doi.org/10.1055/s-1999-6816

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