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Severe mediastinitis caused by an infected bronchogenic cyst

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SUMMARY

Bronchogenic cysts (BCs) are congenital foregut malformations and usually asymptomatic, thin-walled, incidentally diagnosed cysts which can be easily resected by a minimal invasive approach at this time point. However, they may develop symptoms such as infection, bleeding or compression of adjacent structures. There is no consensus about the risk of developing complications during a lifetime; however, recent reports suggest a higher incidence than initially believed. Here, we report a case of severe life-threatening mediastinitis emerging from an infected BC requiring complex surgery, which could have been avoided if surgery had been performed at an early, asymptomatic stage.

BACKGROUND

Bronchogenic cysts (BCs) are rare congenital malformations arising during development of the embryonic foregut and represent the most frequent primary malformations of the mediastinum.[1–3] They are usually located in the mediastinum but can be situated intrapulmonary in 15%–25% of cases.[1 4 5] The majority of BC are diagnosed incidentally; however, symptoms may occur during a lifetime in the context of BC-related complications such as infection, haemorrhage or local extension and compression of adjacent structures. Associated malignancy has also rarely been reported.[2]

It is generally agreed that symptomatic or complicated cysts should be completely resected.[2 4] However, the management of asymptomatic BC remains controversial despite growing evidence that BC-related complications are probably more common and severe than initially anticipated.[2 6] While asymptomatic, thin-walled BC can be easily resected by use of video-assisted thoracic surgery (VATS), whereas surgery for complicated BC may be more complex and less likely achieved by VATS. Here we report a case of severe mediastinitis related to an infected BC localised in the aortopulmonary window which required urgent and extensive surgery in order to control infection. This case report supports existing literature that BC should be resected with minimally invasive techniques before major surgery is required to treat BC-related life-threatening complications.[1–3 6–8]

CASE PRESENTATION

A male patient with a history of smoking and metabolic syndrome was presented to the emergency department of our institution for dyspnoea associated with chest pain, fever and moderate haemoptysis. Clinical examination was unremarkable and laboratory investigations revealed a leucocytosis of 18.3 g/L and elevated C reactive protein and procalcitonin levels of 66.1 mg/L and 5 µg/L, respectively.

INVESTIGATIONS

A chest CT scan revealed a 70×90×110 mm sized, heterogeneous lesion located in the left anterior mediastinum with extensive adjacent mediastinal fat infiltration and pneumomediastinum, suggesting the presence of mediastinitis (figure 1). A thorough anamnesis revealed a pulmonary infection treated with antibiotics almost 20 years ago which, on research, turned out to be related to an infected BC located in the aortopulmonary window.

A transthoracic echocardiogram revealed an extracardiac mass with signs of compression of the left ventricle. Bronchoscopy was without pathological findings, and bronchoalveolar lavage was negative for infection or malignancy.

TREATMENT

An empirical intravenous antibiotic treatment was initiated. However, the evolution was marked by the presence of ongoing mediastinitis with persistent dyspnoea, fever, oxygen dependence, elevation of inflammatory biomarkers, and the development of confusion and hallucinations. As a consequence, surgical debridement of the involved mediastinum with resection of the cyst was performed. Taking into consideration the size of the cyst and the extension of mediastinitis, an open approach via a left hemiclamshell incision was performed.

OUTCOME AND FOLLOW-UP

The postoperative course was uneventful with removal of chest tubes and hospital discharge on postoperative days 4 and 7, respectively. The patient was then transferred to a pulmonary rehabilitation clinic. He was followed up for 3 months postoperatively and presented an uneventful recovery. The histological examination of the surgical specimen confirmed the diagnosis of purulent mediastinitis related to an infected BC arising from the aortopulmonary window (figure 2).

DISCUSSION

Due to the wide use of radiological imaging, an increasing number of BC have been diagnosed in recent years.[3 6] Chest CT scans usually reveal a thin-walled cyst adjacent to the tracheobronchial tree in the upper mediastinum and occasionally in the lung, and usually offer sufficient information for the diagnosis and treatment of BC. However, BC may contain protein-rich liquid which may mimic

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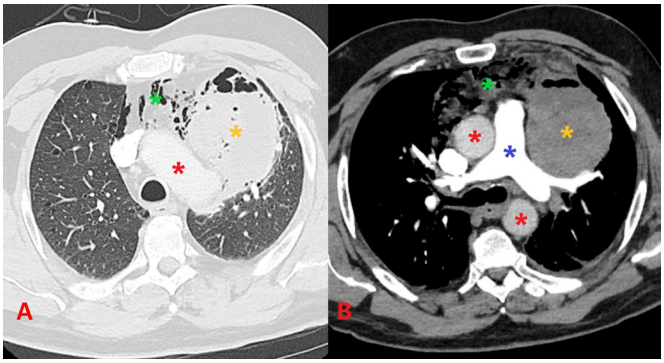

Figure 1 Chest CT scan revealing a large, heterogeneous lesion emerging from the aortopulmonary window (yellow asterisk) associated with pneumomediastinum and extensive mediastinal infiltration, suggesting severe mediastinitis (green asterisk). Note the proximity of the lesion to the aorta (red asterisk) and the pulmonary trunk (blue asterisk).

soft tissue masses in 50% of patients.[2] In these situations, MRI may help to differentiate BC from solid mediastinal masses.[2 3] Nevertheless, a final diagnosis can only be established with histological examination following surgical resection. Some reports have suggested the use of transbronchial needle biopsy and aspiration of the BC content for diagnosis and treatment of BC.[3 6 7] However, it has been shown, that this approach usually leads to recurrence since the secretion-producing mucosa remains in place. Moreover, there is a risk of subsequent BC infection due to inoculation of germs during biopsy.[3]

The treatment of BC remains controversial to date. While most authors agree that surgical resection is indicated for symptomatic or complicated BC,[1–4] the management of incidentally discovered asymptomatic BC remains controversial. However, there is growing evidence that the evolution of BC may be unpredictable and can lead to complications which may be life-threatening, including severe haemoptysis, haemothorax and tension pneumothorax, as well as compression of surrounding organs leading to cardiac tamponade, arrhythmia and superior vena cava syndrome.[1–5]

Kirmani *et al* performed a review of the literature including 23 reports with 683 patients in order to investigate the need for surgery for asymptomatic BC. They identified a group of 74 asymptomatic patients undergoing follow-up without surgery, in which 33 (45%) subsequently developed symptoms requiring resection. In 5 of 683 patients (0.7%), the surgical specimen revealed malignancy.[6] In addition, several reports indicate significantly more surgery-related complications and longer hospital stays following resection of symptomatic or complicated BC when compared with asymptomatic and non-complicated BC.[1–3 6] The same holds true for resection of BC larger than 5 cm, a size cut-off which seems to be associated with increased operating time, blood loss and surgical complexity.[2] For those reasons, there is growing evidence in recent literature that surgery should be performed even in asymptomatic, incidentally diagnosed BC.[1–3 6]

Complete resection of BC is mandatory in order to remove all remaining mucosa and thus decrease the risk of recurrence and complications.[1–3] In the case of an uncomplete resection due to technical issues, de-epithelisation with cauterisation of the remaining mucosa is recommended.[2 3] Recent reports suggest that a minimally invasive surgical approach such as VATS[1–3 7] or robotic-assisted surgery[8] is appropriate for the majority of BC and seems to be associated with less postoperative pain and length of stay and chest tube drainage compared with open surgery without increasing the risk of incomplete resection or perioperative complications.[7] However, large or complicated BC may still require complex surgery and an open approach in order to prevent intraoperative complications or an incomplete resection.

This report of severe mediastinitis related to an infected BC, adds another piece of evidence that BC should be resected with minimally invasive techniques before major surgery is required to treat BC-related life-threatening complications.

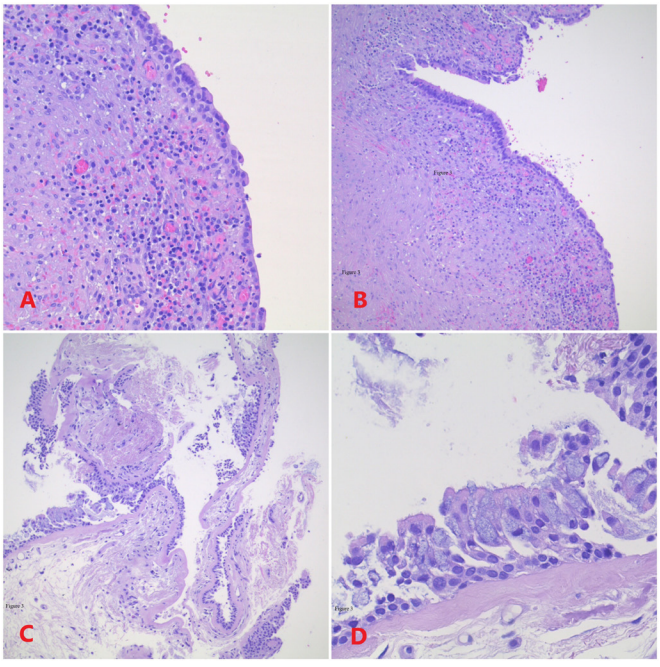

Figure 2 Histological assessment of the surgical specimen with the presence of a BC, lined by a ciliated columnar epithelium (A,B), with thickened cystic wall by a reactive myofibroblastic proliferation with presence of reactive atypia accompanied with a mixed inflammatory mononucleated and multinucleated infiltrate (C,D). BC, bronchogenic cyst.

Learning points

- Bronchogenic cysts (BCs) are congenital foregut malformations which are usually asymptomatic. When incidentally discovered, they can be easily resected by a minimally invasive approach.
- BCs tend to grow over time and symptoms may appear in the context of cyst-related complications such as infection, haemorrhage or local extension and compression of adjacent structures. Associated malignancy has also been rarely reported.
- Complicated BCs could be challenging to manage and usually require more extensive surgery with a higher rate of postoperative complications.
- We encourage an upfront surgical treatment of even asymptomatic BCs using minimally invasive techniques when possible.

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REFERENCES

1 Ceylan KC, Batihan G, Üçvet A, *et al*. Surgery in congenital lung malformations: the evolution from thoracotomy to VATS, 10-year experience in a single center. *J Cardiothorac Surg* 2021;16:131.
2 Wang X, Li Y, Chen K, *et al*. Clinical characteristics and management of primary mediastinal cysts: a single-center experience. *Thorac Cancer* 2020;11:2449–56.
3 Aker C, Sezen CB, Dogru MV, *et al*. Prognostic factors and long-term results in patients who underwent Videothoracoscopic bronchogenic cyst excision. *Ann Thorac Cardiovasc Surg* 2021;27:225–9.
4 Lardinois D, Gugger M, Ris HB. Bronchogenic cyst of the left lower lobe associated with severe hemoptysis. *Eur J Cardiothorac Surg* 1999;16:382–3.
5 Bayfield N, Stamp N, Laycock A, *et al*. Large air-filled intrapulmonary bronchogenic cyst associated with tension pneumothorax during air travel. *BMJ Case Rep* 2019;12:e228032.
6 Kirmani B, Kirmani B, Sogliani F. Should asymptomatic bronchogenic cysts in adults be treated conservatively or with surgery? *Interact Cardiovasc Thorac Surg* 2010;11:649–59.
7 Guo C, Mei J, Liu C, *et al*. Video-Assisted thoracic surgery compared with posterolateral thoracotomy for mediastinal bronchogenic cysts in adult patients. *J Thorac Dis* 2016;8:2504–11.
8 Chen K, Zhang X, Jin R, *et al*. Robot-Assisted thoracoscopic surgery for mediastinal masses: a single-institution experience. *J Thorac Dis* 2020;12:105–13.