

CASE REPORT

Lung-sparing approach for an intrapulmonary bronchogenic cyst involving the right upper and middle lobes

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SUMMARY

Intrapulmonary bronchogenic cysts (IBC) represent 20% of abnormal budding of the respiratory tract. Lobectomy is the recommended treatment for IBC in symptomatic adults. We presented a case of a patient with an IBC involving the right upper and middle lobes (RUL–RML). A 27-year-old woman presented with a 2-month history of thoracic pain, cough and haemoptysis. An opacity was found on the chest X-ray. High-resolution CT/MRI showed a 7×4.5 cm marginated mass with an air bubble inside. A video-assisted thoracoscopic surgery was performed. The cyst was neither palpable nor visible. An intraoperative ultrasonography localised the cyst involving the RUL–RML. The lung above the cyst was incised, and a greenish-mucoid content was aspirated. A branch of the superior pulmonary vein was visible. The remaining cystic wall was cauterised. The patient was discharged on day 4. Histology confirmed the IBC. The patient is asymptomatic at a 16-month follow-up. The lung-sparing operation in a young woman with IBC involving the RUL–RML has been beneficial. A long-term follow-up is mandatory.

BACKGROUND

Intrapulmonary bronchogenic cysts (IBC) represent about 20% of bronchogenic cysts (BC). Most undiagnosed IBC in neonatal age become symptomatic in adulthood. Lobectomy is the recommended treatment for IBC in symptomatic adults.

We presented a young patient with an IBC involving the right upper and middle lobes (RUL–RML) successfully treated with a video-assisted thoracoscopic surgery (VATS) lung-sparing approach, which was preferred to the more aggressive traditional treatment.

CASE PRESENTATION

A 27-year-old woman presented with a 2-month history of thoracic pain, cough and haemoptysis. Her medical history presented only a respiratory distress in early childhood.

INVESTIGATIONS

The chest X-ray demonstrated an opacity in the right middle field. A high-resolution CT scan showed a marginated mass (7×4.5 cm) in the anterior segment of RUL with an air bubble inside, polylobulated margins and adherent to the superior pulmonary vein (SPV; figure 1A). It also revealed a hypoperfused area in the contiguous parenchyma with an endoluminal defect of the pulmonary vein

branch for the anterior segment of RUL. MRI confirmed this finding, consistent with a cystic lesion containing dense fluid. The described mass was in contact with the upper and lower pulmonary vein. Bronchoscopy was negative as well as cytological and microbiological analysis.

TREATMENT

A right VATS was performed. The whole lung was adherent to the chest wall, and therefore a mini-thoracotomy was necessary. There was no horizontal fissure and the cyst was neither visible nor palpable. An intraoperative ultrasonography allowed to localise the cyst with liquid content, involving the RUL–RML, that appeared adherent to the SPV. This was confirmed by fine needle aspiration that showed a greenish and thick fluid. A bilobectomy was considered too aggressive so a conservative approach was preferred. The lung above the cyst was incised towards the SPV. The cyst was opened and a greenish mucoid content was aspirated. After removing the anterior cystic wall, exploration of the cyst revealed a branch of the SPV (figure 1B). The remaining portion of the wall was carefully cauterised, and lavage with iodinated polyvinyl polymer was also performed. Two different bioptic specimens were taken for diagnostic and comparative purposes: one from the RUL (hypoperfused) and the other one from the right lower lobe (normally perfused). Finally, two chest drains were placed on suction, one of them inside the remaining cystic cavity. No air leakage was noted.

OUTCOME AND FOLLOW-UP

Histology confirmed the IBC. Microscopic specimens: vessels with thickened wall were found in the parenchymal specimens from both lobes (figure 2A); bronchial-type epithelium and hyaline cartilage plaques appeared in the cystic wall (figure 2B).

Postoperative course was uneventful and the patient was discharged on day 4. The patient is asymptomatic with no recurrence at a 16-month follow-up.

DISCUSSION

BCs are congenital lung anomalies resulting from an abnormal budding of the respiratory tract, and IBC represent about 20% of these lesions. They can be found either in the mediastinum or within the pulmonary parenchyma. When these abnormal buddings occur during early development, the cyst

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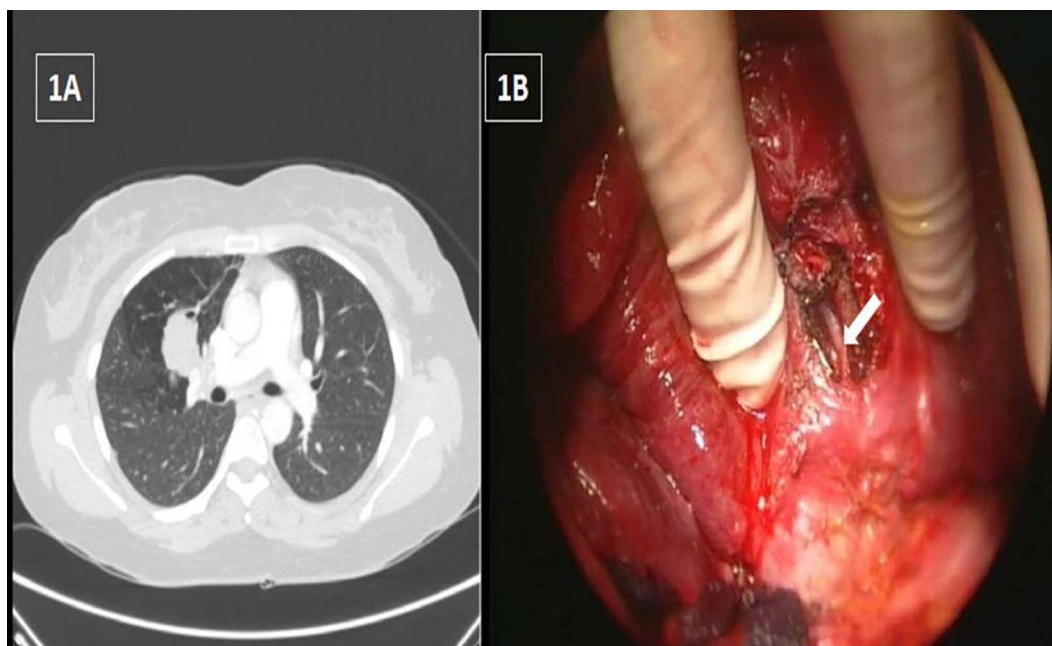


Figure 1 (A) High-resolution CT/MRI showing a 7×4.5 cm margined mass with an air bubble inside, and a hypoperfused area in the distal parenchyma. (B) After removing part of the cystic wall, a branch of superior pulmonary vein was visible (arrow).

tends to be located along the tracheobronchial tree and in the mediastinum, while cysts that arise later are more peripheral and may be located within the lung parenchyma.

Although most authors report that IBC are often asymptomatic, Kirmania *et al*¹ have demonstrated that only 74 (10.8%) out of 683 patients with BC are asymptomatic; of those patients 33 (45%) eventually developed symptoms requiring surgery. The presenting symptoms can be manifestations of the compression of major airways by the cyst, infection of the cyst and pneumonia in the adjacent parenchyma. Reported complications include superior vena cava syndrome, tracheal compression, pneumothorax, pneumonia, pleural effusion, pericardial tamponade, carcinomatous or sarcomatous transformation.^{2–6} The gold standard classically considered is the complete resection of the cyst, and segmental or lobar resections are required procedures.⁷ Moreover, surgical excision is often the only method that allows to establish the definitive histopathological diagnosis.

There have been many reported cases in which surgical resection is difficult due to the presence of dense pericystic adhesions

to adjacent structures, and just a partial resection of the cyst is possible.⁷ In these cases, BC are drained or partially excised, and long-term follow-up is indicated to detect cyst recurrences.^{8–9} Because of this, surgical excision is recommended even for asymptomatic cysts, to prevent complications and operative difficulties.^{10–11} We agree with several authors that VATS lung-sparing technique is valuable as a lung preservation strategy, especially for asymptomatic patients.^{12–13}

To answer the old question: should patients with asymptomatic IBC be operated? A recent report shows that early surgical resection of BCs provides better conservation of pulmonary parenchyma and a lower incidence of inflammatory lesions which are present in 84% of adults.¹⁴

We agree that asymptomatic patients should be informed about the 20% morbidity of surgery, the 45% risk of developing symptoms and finally of the 0.7% risk of malignancy. When patients choose non surgical management, long-term follow-up is mandatory.¹ When patients choose non surgical management, long-term follow-up is mandatory (1).

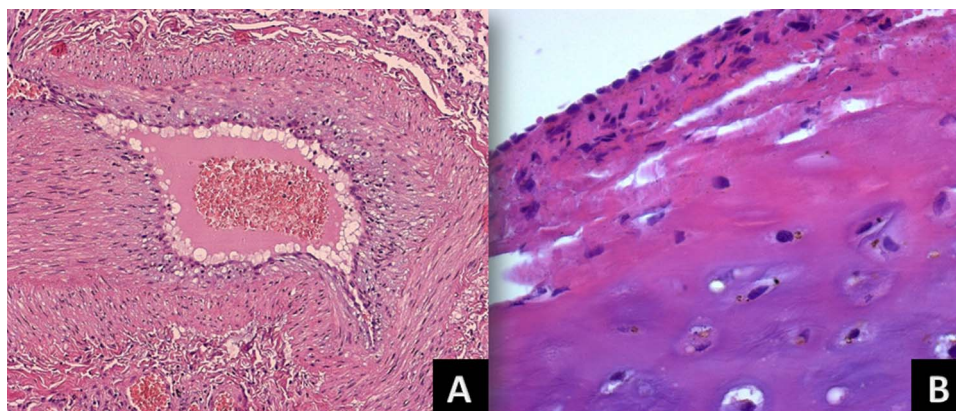


Figure 2 (A) An intraparenchymal vessel with thickened walls (H&E ×200). (B) Bronchial-type epithelium and cartilage are apparent in this bronchogenic cyst (H&E ×400).

The VATS lung-sparing approach performed in a young woman with an IBC involving RUL–RML has been beneficial, and moreover a bilobectomy has been avoided.

Learning points

- ▶ Surgical excision for asymptomatic intrapulmonary bronchogenic cysts (IBC) is recommended to prevent complications and operative difficulties.
- ▶ A video-assisted thoracoscopic surgery lung-sparing approach is feasible and should be offered to patients with IBC.
- ▶ Long-term follow-up is mandatory to detect recurrences and malignant transformation.
- ▶ When the patient opts for conservative management, close long-term follow-up must be guaranteed.

Contributors AC and AS conceived and drafted the manuscript. DC helped with the draft. MM performed the operation and co-ordinated the study. All authors read and approved the final manuscript.

Competing interests None.

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