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Intrapericardial Development of a Bronchogenic Cyst – Case Report

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Rezumat

Dezvoltare intrapericardică a unui chist bronhogenetic – prezentare de caz

Introducere: Chisturile bronhogenetice, localizate frecvent la nivelul mediastinului sau la nivelul parenchimului pulmonar, iau naștere prin înmugurirea anormală a arborelui traheobronșic. Având în vedere că există un număr necunoscut de pacienți asimptomatici ce rămân nediagnosticați, adevărata istorie naturală a acestor adulți este necunoscută și toate studiile publicate suferă din acest punct de vedere – sunt astfel selectați doar pacienții simptomatici și unii pacienți asimptomatici.
Case report: Prezentăm cazul unei paciente de 42 de ani care s-a adresat serviciului nostru pentru o durere toracică localizată central, care s-a înrăutățit pe parcursul a mai multe săptămâni. După efectuarea unui set de investigații (hemoleucogramă, biochimie, radiografie toracică standard, electrocardiogramă, examen CT cu substanță de contrast administrată intravenos), am identificat o masă mediastinală situată inferior de carena traheală și adiacentă la pleura stângă, atriul stâng, artera pulmonară și esofag. Având în vedere dimensiunile acestei formațiuni, s-a propus și efectuat intervenția chirurgicală. În cursul acesteia am reușit să îndepărtăm complet chistul, iar pacienta a avut o evoluție postoperatorie fără evenimente.
Discuții: Un chist bronhogenetic poate fi dificil de diferențiat de alte leziuni (alte leziuni chistice dobândite, mase mediastinale). Complicații serioase ale chisturilor bronhogenetice sunt rare, acestea incluzând: sindrom de venă cavă superioară, compresiune traheală, pneumotorax, pleurezie și pneumonie. Excizia chirurgicală este tratamentul recomandat, chiar și pentru chisturile asimptomatice, pentru a preveni complicațiile și incidentele intraoperatorii.
Concluzii: Chisturile bronhogenetice sunt rare și de obicei asimptomatice. Pacienții asimptomatici pot deveni simptomatici în orice moment și pot dezvolta complicații amenințătoare de viață. Diagnosticul definitiv este de obicei obținut după rezecția chirurgicală completă, la examenul anatomopatologic. Bazându-ne pe experiența noastră și studiind datele din literatură, putem concluziona că chisturile bronhogenetice trebuie tratate chirurgical atunci când sunt identificate, iar tratamentul conservator nu este recomandat.

Cuvinte cheie: chist bronhogenetic, intervenție chirurgicală, complicații

Abstract

Introduction: Bronchogenic cysts, commonly located in the mediastinum or lung parenchyma, arise from abnormal budding of the tracheobronchial tree. Since an unknown percentage of asymptomatic adult patients with bronchogenic cysts remain undiagnosed, the true natural history ofthese cysts in adults is uncertain and the available series reflect a significant selection bias in favor of the symptomatic individual.
Case report: We present the case of a 42-year-old female who presented with adull central chest pain, which gradually worsened overseveral weeks. After several tests performed (standard blood tests, standard X-Ray, ECG, angio-CT scan),

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we identified a mediastinal mass inferior to the carina and adjacent tothe left pleura, left atrium, pulmonary artery and esophagus. Considering the size of this mass and the possible complications, surgery was proposed and preformed. We were able to completely remove the cyst and the patient had no complications after surgery.

Discussions: It may be difficult to differentiate a bronchogenic cyst from other lesions (acquired cystic lesions, mediastinal masses). Serious complications from bronchogenic cysts are rare, but can include SVC syndrome, tracheal compression, pneumothorax, pleurisy and pneumonia. Surgical excision is recommended even for asymptomatic cysts, to prevent complications and operative difficulties.

Conclusions: Bronchogenic cysts are rare and usually asymptomatic. Asymptomatic patients with bronchogenic cysts may become symptomatic cases, and in time may develop life-threating complications. Definitive tissue diagnosis is usually available only after surgical excision. Based on our experience and after studying data from literature, we can conclude that bronchogenic cysts should be treated surgically and that a conservative approach is not recommended.

Key words: bronchogenic cyst, surgical intervention, complications

Introduction

Bronchogenic cysts, commonly located in the mediastinum or lung parenchyma, arise from abnormal budding of the primitive tracheobronchial tube between the 26th and 40th days of gestation (1, 2). The location of the cyst depends on the embryological stage of development at which the anomaly occurs; the cyst tends to be located along the tracheobronchial tree when this anomaly occurs earlier and within the lung parenchyma when it occurs later (3, 4). Bronchogenic cysts are a rare cause of mediastinal mass (4, 5). Bronchogenic cysts are typically asymptomatic, with many detected incidentally. An unknown percentage of asymptomatic adult patients with bronchogenic cysts remain undiagnosed and the true natural history of these cysts in adults is uncertain.Interpretation of the available literature is complicated by diagnostic uncertainty in the absenceof surgical resection for a definitive diagnosis(6). However, complications do occur and some other times differential diagnosis is needed. Resection of even the asymptomatic cyst is recommended because of the possibility of complications and the difficulty of operating on complicated cysts (6, 7).

Case report

A42-year-old female presented with a dull central chest pain, which gradually worsened overseveral weeks. At first the patient was admitted in Cardiology, where a standard chest X-ray showed a mediastinal mass. The standard blood tests were within normal limits, with no markers of myocardial ischemia. The ECG was also normal. The investigations were continued with a chest angio-CT which showed a 9 cm posteriormediastinal mass inferior to the carina and adjacent tothe left pleura, left atrium, pulmonary artery and esophagus (Fig. 1).

There was marked mass effect with leftatrium and superior vena cava (SVC) compression and rightinferior pulmonary vein congestion (Fig. 2).

Clinical assessment did not reveal evidence of superior vena cava obstruction or hemodynamic compromise. Transthoracic echocardiography revealedthe mass as separate from the left atrium, howeverfurther comment on likely diagnosis was not possiblefrom the views obtained, raising the question if this mass was an atrial mixoma or something else (lymphoma, bronchogenic cyst). At this time the patient was referred to our department for further management.

After reviewing the patient's data, we decided to operate her. Upon thoracotomy the mass was identified and, after removing the adhesions with the pericardium, esophagus, inferior trachea and mainstem bronchi, the mass was punctured with a fine needle (Fig. 3 A).

A clear liquid was aspired and we decided to continue evacuating the cyst (Fig. 3 B). After the cyst was completely evacuated, its walls were sectioned. At this moment it became clear that the cyst had an intrapericardial development, as we

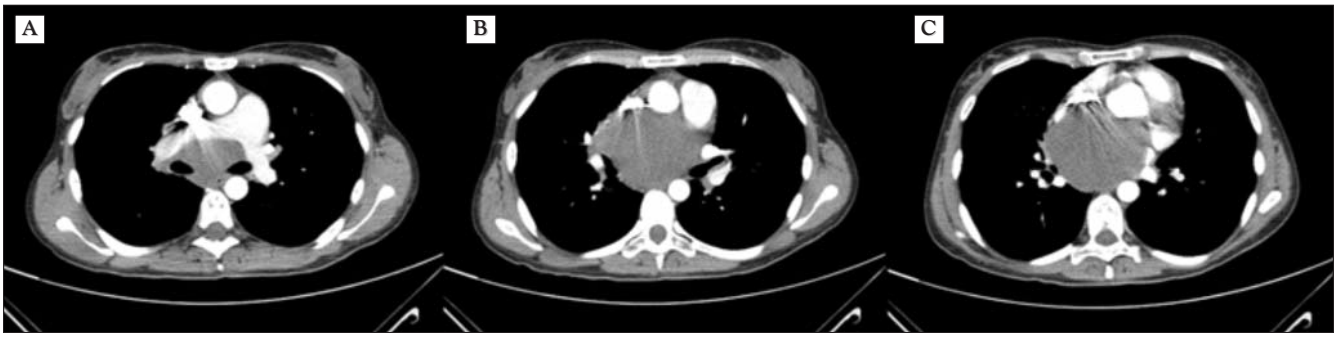

Figure 1. *Angio-CT aspects of a mediastinal mass, with its upper limit inferior to carina (**A**), going down with a mass effect on the heart (**B** and **C**).*

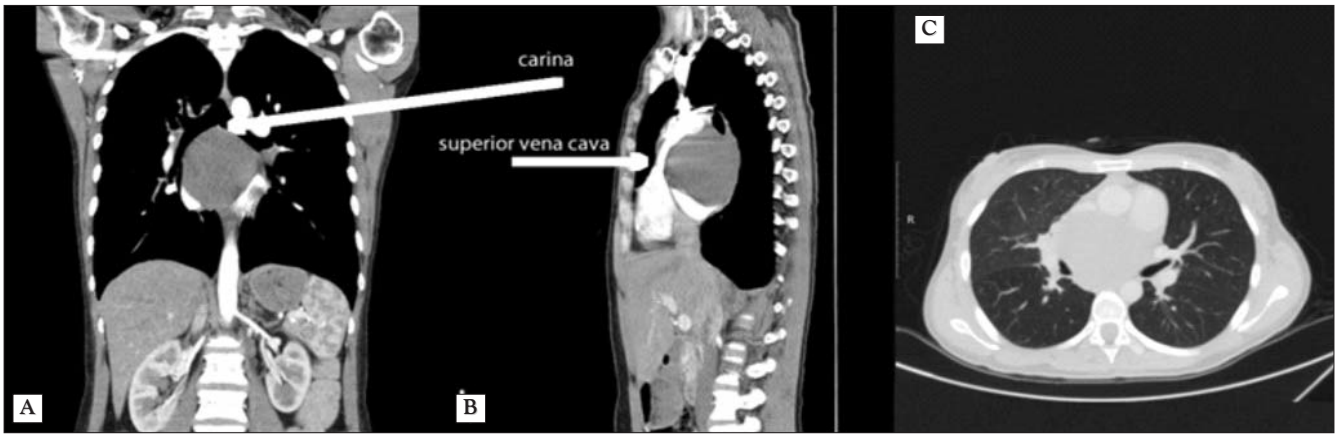

Figure 2. *Angio-CT aspects of a mediastinal mass, with its upper limit inferior to carina (**A**), compressing the superior vena cava (**B**); detail of lung parenchyma (**C**).*

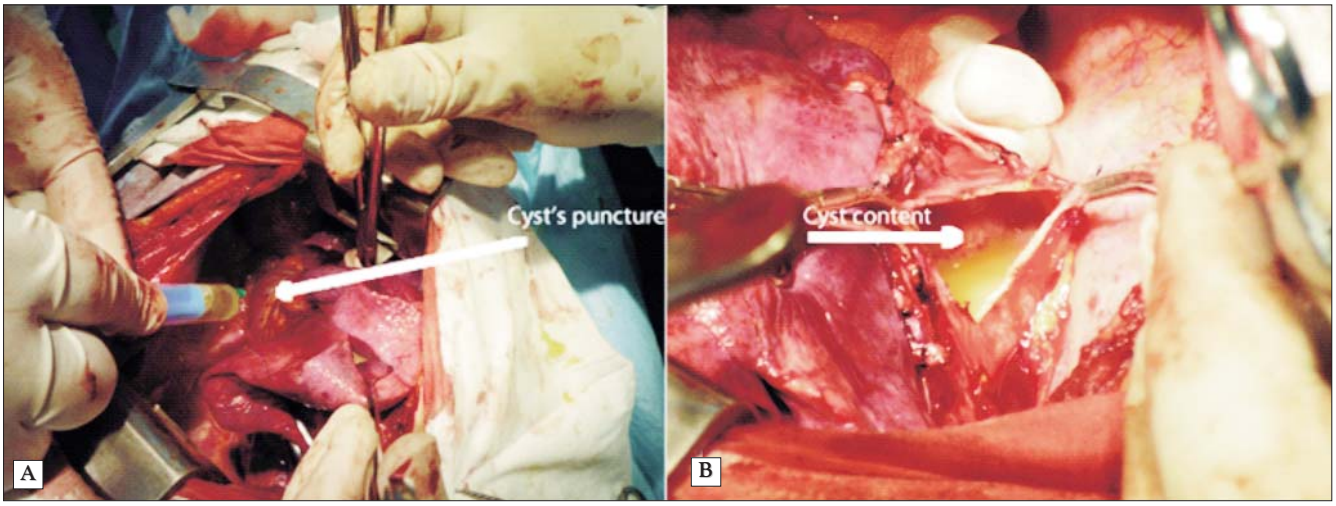

Figure 3. *Intraoperative aspects – (**A**) puncture of the cyst revealing its content, (**B**) an incision was performed in order to inspect the cyst's interior and content.*

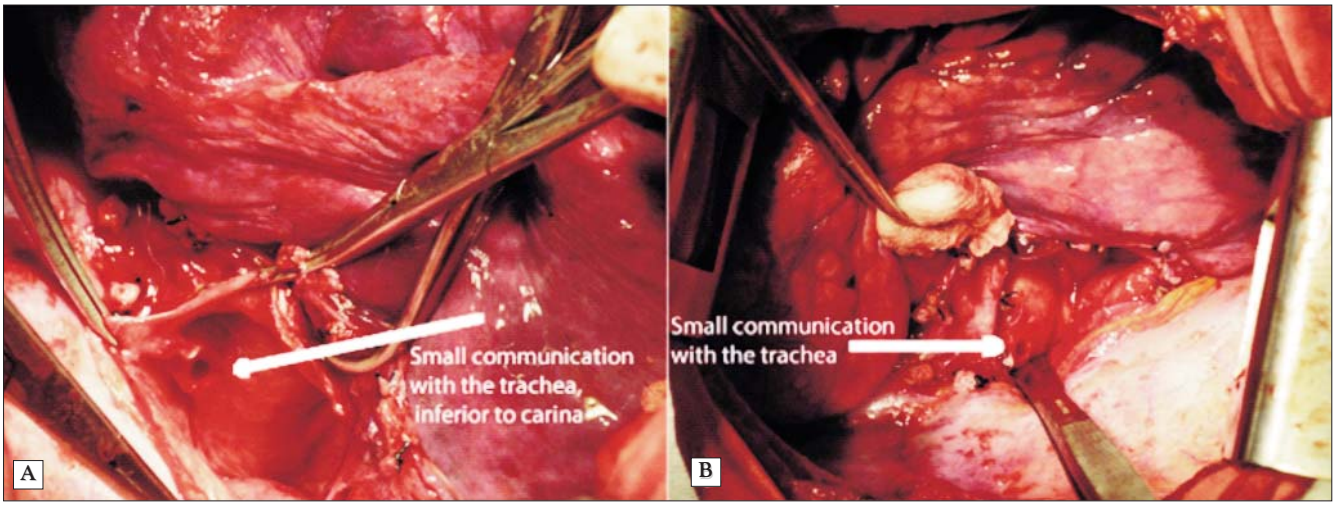

Figure 4. *Intraoperative aspects of the cyst, revealing a small communication with the trachea*

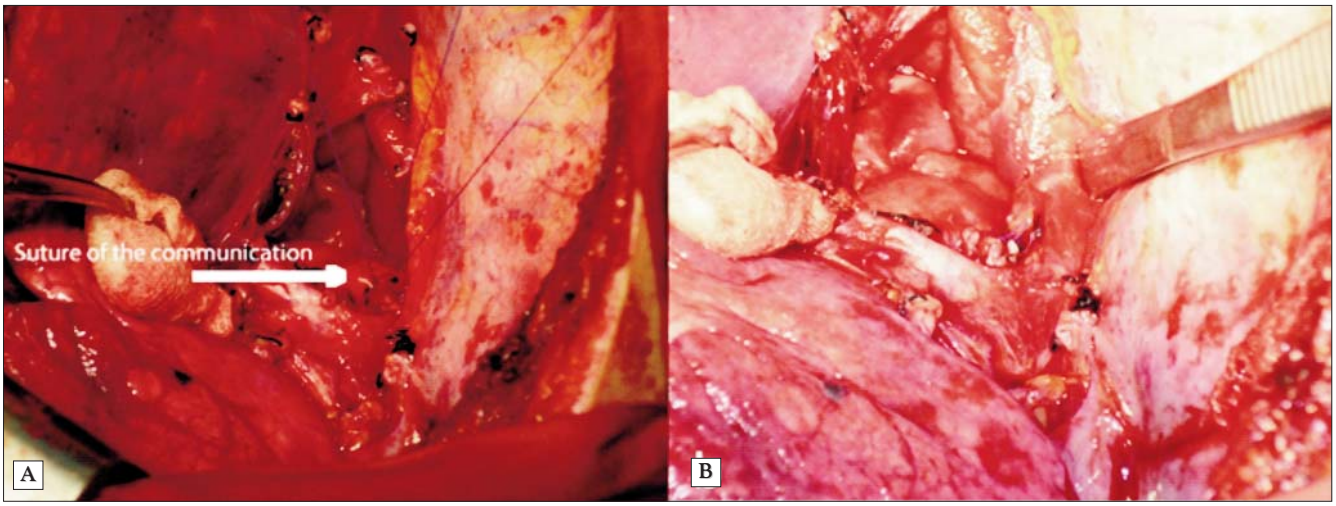

Figure 5. *Intraoperative aspects – (**A**) suture of the communication; (**B**) final intraoperative aspect.*

could see that upon sectioning the cyst's wall, the pericardial sac has been opened.The diagnosis of an intrapericardial cyst was based on the fact that there was no pericardium between the cyst and the region of the superior vena cava, the pulmonary artery and the aorta. Also, a possible small communication with the trachea was identified - suggesting its origin from the anterolateral wall of the trachea (Fig. 4 A, B).

This communication was sutured because of the high risk of a fistula (Fig. 5 A, B).

A part of the cyst wall was left attached to the great vascular structures due to extensive pericystic fibrotic adhesions. Postoperatively, the patient was anti-coagulated with a standard regimen of low molecular weight heparin. There were no complications after surgery, the patient being discharged on the 7th day after the surgical intervention.

Discussions

It may be difficult to differentiate a bronchogenic cyst from other lesions (acquired cystic lesions, mediastinal masses). Because there are no specific CT or MR imaging findings that allow easy differentiation, reviewing the patient's clinical history and previous radiologic studies may be helpful (8). While the anatomy of the mediastinal mass is easily seen using a CT scan, the modality is limited in its ability to diagnose bronchogenic cysts of high density. The attenuation of the cyst's contents can vary from that of water to soft-tissue (9, 10). The value of attenuation can be as high as 100 Hounsfield units if the cyst contains protein or calcium oxalate (10). In this case the CT scan revealed a fluid level of attenuation.

Serious complications from bronchogenic cysts are rare, but can include SVC syndrome, tracheal compression, pneumothorax, pleurisy and pneumonia (10). Fortunately our patient did not suffer from any of these. Atrial fibrillation secondary to bronchogenic cyst has also been described (12).

Bronchogenic cysts are found most frequently along the tracheobronchial tree in the mediastinum or within the lung parenchyma. Rarely, the cysts have occurred in other locations, including cutaneous (13) and subcutaneous tissues (14), neck (15), pericardium (16), diaphragm (17), abdomen (18), and the intramedullar part of the spine (19). They have also been reported to extend from the mediastinum through the diaphragm into the abdomen as dumbbell cysts (19). Studies by DiLorenzo et al. (3), ST. Georges et al. (4), and Patel et al. (21) indicate that the frequency of mediastinal bronchogenic cysts is greater, whereas other series (11), reflect a higher frequency of cysts with intrapulmonary location (21, 22, 23, and 24).

Surgical excision is recommended even for asymptomatic cysts, to prevent complications and operative difficulties(4, 21, and 25). Although our department has experience with bronchogenic cysts, this is the first intrapericardial bronchogenic cyst diagnosed and treated in our service. The specific literature is abundant with case presentations and case series of bronchogenic cysts, but only a few cases of intrapericardial bronchogenic cysts are published (26, 27), and we haven't found yet any case series regarding the analysis of intrapericardial bronchogenic cysts.

Conclusions

Bronchogenic cysts are a rare, usually asymptomatic condition. Asymptomatic patients with bronchogenic cysts may become symptomatic, and in time may develop life-threating complications (SVC syndrome, tracheal compression, pneumothorax, pleurisy and pneumonia). Definitive tissue diagnosis is usually available only after surgical excision. Based on our experience and after studying data from literature, we consider that bronchogenic cysts should be treated surgically and that a conservative approach is not recommended.

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