Acute respiratory distress due to a bronchogenic cyst exclusively managed with minimally invasive techniques

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Abstract

Mediastinal bronchogenic cysts are usually asymptomatic being often diagnosed in young adults. We present a case of a 2-yearold boy in severe respiratory distress due to a large mediastinal cyst that underwent urgent ultrasound-guided percutaneous decompression followed by planned strictly thoracoscopic excision. This unique report highlights that bronchogenic cysts may develop acute compressive symptoms requiring prompt drainage, which enables elective resection; and that both proceedures can be accomplished successfully by fully minimally invasive techniques.

Introduction

Bronchogenic cysts (BC) are usually asymptomatic and often incidentally diagnosed in older children and adults. However, large mediastinal BC may compress the tracheobronchial tree leading to respiratory manifestations that mimic infection/atelectasis on plain x-ray¹.

Herein, a clinical case of severe respiratory distress due to a mediastinal BC that was managed exclusively by a minimally invasive approach is presented.

Case Report

A 2-year-old boy, with a history of recurrent wheezing, was referred to the pediatric intensive care unit in acute respiratory distress. He was febrile and presented tachypnea, dyspnea, stridor, global chest retraction and hypoxia. Over the last 25 days, he had been admitted twice due to cough, wheeze and mild respiratory distress. Thoracic CT scan revealed a well-circumscribed, bilobed cyst (48x46x30 mm) in the superior and posterior mediastinum, deviating the supra-aortic vessels, and compressing the esophagus and the trachea; there was a cervical extension, contiguous to the left thyroid lobe and common carotid artery (Fig.1A-B).

Towards the severe respiratory distress, in an attempt to stabilize the patient condition and improve ventilation, a decision was made to perform a percutaneous cyst drainage.

Under ultrasound and fluoroscopic guidance, an 8Fr pigtail catheter was inserted into the cyst with aspiration of 40 cc of fluid, which resulted in near complete collapse of the cyst (Fig. 1C) and immediate clinical improvement. Elective thoracoscopic excision of the mass was then planned.

With the patient in modified right lateral decubitus, three 5mm trocars were inserted. The cyst was identified posterior to the left subclavian artery (Fig. 2A-C); because of the intimate relation with the esophageal wall, an intra-operative upper endoscopy was performed to assist the dissection. The cystic mass was then removed *en bloc*, the resulting muscle defect of esophageal wall being closed with interrupted absorbable sutures; a thoracic tube was left in place through one port.

The postoperative course was uneventful. After removal of the thoracic tube, the child was discharged home on postoperative day 1. Histological examination revealed a uniloculated cyst lined by respiratory type epithelium with underlying fascicles of smooth muscle, respiratory-type mucous glands and cartilage, consistent with a bronchogenic cyst.

At 2-year follow-up, the child is doing well with no digestive or respiratory symptoms; the scars are almost imperceptible.

Discussion

Foregut cystic malformations are rare congenital entities. Bronchogenic cysts (BC) in the mediastinum form early in fetal development from abnormal buds of tracheobronchial tree. Most of them are asymptomatic at the early stage, thus they are rare in infancy and often recognized in young adults. In fact, most patients are only diagnosed when the BC become infected or grow large enough to compress adjacent organs¹.

The treatment of asymptomatic BC is not consensual. In adults, conservative management under close long-term follow-up is an option². In children, both symptomatic and asymptomatic cysts should be surgically excised because there is risk of enlargement/compression (due to secreting mucosa or infection), erosion/perforation, bleeding, and malignant degeneration. As these lesions do not regress, it is probably more appropriate to resect asymptomatic BC not only in children but also in the young and in the healthy adult in order to prevent life threatening complications such as the present case illustrates ¹.

Drainage of symptomatic BC is a minimally invasive procedure when performed by percutaneous access under ultrasound guidance or by transesophageal endoscopic ultrasonography ³. Drainage can be used for decompression in patients who are nonsurgical candidates, but it is less than optimal definitive treatment because leaves the risk of cyst and/or infection recurrence. Beyond the rapid resolution of respiratory distress with marked improvement in ventilation, the drainage may be viewed as a first step to facilitate the subsequent cyst excision due to reduction of size and inflammation³.

Surgical excision of mediastinal BC is demanding because of vicinity or even adhesions to surrounding vital organs. Moreover, as some patients do not tolerate thoracoscopy well, thoracotomy is the procedure usually performed such as we have found in the reports of patients requiring decompression drainage ³. Video-assisted thoracoscopic surgery (VATS) has been increasingly used to excise BC and is the preferred choice of most thoracic surgeons ⁴. However, a strictly thoracoscopic procedure, as the present one, is viable despite the tigh space available, mainly for suturing. So, if the patient tolerates, thoracoscopy should be endeavored. It is challenging because involves the use of advanced minimally techniques such that a high level of surgical skills.

In conclusion, this unique report demonstrates the feasibility of strict and fully minimally invasive techniques to decompress and to excise a BC, which were crucial for the excellent outcome.

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Figure legends

Figure 1 . A-B: CT scan before drainage; C: MRI after drainage.

Abbreviations : Ao- aortic arch; BC- bronchogenic cyst; Eso- esophagus; LcCA- left common carotid artery; LSA- left subclavian artery; Tra- trachea

Figure 2 . A-C: Intraoperative view.

Abbreviations : Ao- aortic arch; BC- bronchogenic cyst; Eso- esophagus; LSA- left subclavian artery; LSV- left subclavian vein; PhN- phrenic nerve



