

Case report

Tracheal hamartoma: pericardial flap replacement of membranous tracheal wall

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Abstract

We presented a rare case of large tracheal hamartoma in a 14-year-old boy and its management with a novel technique of reparation with pericardium of entire membranous portion of the trachea, after the failure of more conservative approaches like bronchoscopic resection and stenting. We remark the advantages of autologous material instead of prosthetic material. © 2002 Elsevier Science B.V. All rights reserved.

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1. Case report

A 14-year-old boy was admitted to our hospital for severe dyspnea due to major airway obstruction. In previous 6 months he was attended for two episodes of mild respiratory obstruction diagnosed as bronchial asthma. Superior mediastinal enlargement evidenced by chest X-ray led to a more detailed study. Helical CT scan showed a posterior mediastinal tumor that invaded the membranous portion of the trachea (MPT) from cricoid to 1 cm above the carina (Fig. 1A). The first attempt was rigid bronchoscopic resection and airway control with nasotracheal tube. Histopathologic analysis resulted a hamartoma. After 2 days a dynamic stent (Rusch, Kern, Germany) was placed and the patient discharged to pediatric unit (Fig. 1B). He was newly admitted 2 weeks later due to progressive respiratory difficulty; bronchoscopy evidenced tumoral protrusion of posterior wall of the stent with an important narrowing. The repeated biopsy confirmed the hamartoma as we present on Fig. 1C.

With institutional approval and informed consent, a combined cervico-external approach was performed, exposing the mediastinum and trachea. During all operation the ventilation was maintained through orotracheal tube using the stent to bypass the temporal tracheal defect, avoiding replacement it. A large tumor was identified emerging of MPT, without compromise of oesophagus or other mediast-

inal structure. Resection included 8 cm of MPT, preserving the cartilages and the left arterial irrigation.

A pericardial flap was raised, keeping its attachment to the aortic root and passed under brachiocephalic arterial trunk to the right side of the trachea. It was sutured in a continuous fashion with absorbable material (Biosyn 4/0, AutoSuture, USA) like a new posterior wall, to both cartilaginous edges (Fig. 2, left). The patient was successfully extubated at the end of the operation. The post operative period was free of morbidity. The stent was left in place and withdrawn 4 weeks later.

There was not recurrence at 10th month of follow up and complete relieve of dyspnea was achieved. Flexible bronchoscopy showed a deformity of right lateral wall without narrowing the trachea (Fig. 2B,C). The vocal cord mobility and voice were normal.

2. Discussion

Few cases of tracheal hamartoma (THAM) have been published in English literature and only two cases are children [1,2]. There is no other report of extensive compromise of the airway by this type of tumor.

Hamartoma is a benign disease characterized by a disarray of normal local tissues and is formed exclusively by mesodermal derivatives. A total of 90% of lung hamartomas are located into the pulmonary parenchyma and occasionally are found in young people. Resection is the definitive treatment and the recurrence is extremely rare [3]. As other authors refer [1,2,4], our patient presented an atypical

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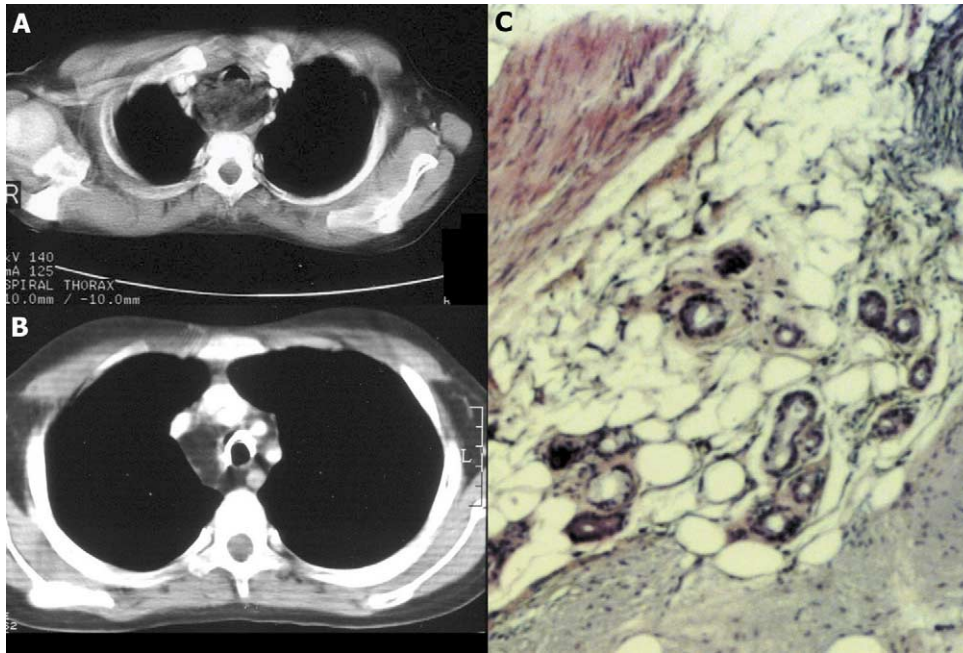


Fig. 1. CT scan shows a Hamartoma that compromises the posterior membranous portion of the trachea (A). Dynamic stent is showed in place (B). Histology slide of the tumor with a mixture of mature cartilage, fibrous, muscle, and adipose tissue (C).

history of respiratory difficulty, treated initially as asthma and bronchial infection. These history findings are similar with other tracheal tumors that growth slowly into the airway, and remark the high index of suspicion necessary to detect them. Helical CT scan and bronchoscopy are valuable methods to confirm the diagnosis of tracheal tumor and to provide detailed information about size, extension and etiology. Repeated biopsies were required to establish the histology of the tumor because its unusual location. Differential diagnosis includes teratoma [2], a far more frequent cause of mediastinal tumor than THAM. Its origin

from MPT was established by its transmural compromise, benignity and one mass appearance.

Our decision to operate the patient on was based on the benign character of THAM, the patient's age and by the failure of less invasive treatments.

It is difficult to reach a definitive solution in patients that require a wide tracheal resection. Today, there is not any material able to resemble the structure and properties of tracheal wall, and when is possible, resection with primary end to end anastomosis should be performed. Although prosthetics materials can be used to repair tracheal defects, autologous tissues avoid many troubles like excessive inflammatory response, rejection, infection, dehiscence and migration. Anterior patch tracheoplasty (APT), described in the management of congenital-complete tracheal ring, uses free pericardium to enlarge the cross sectional diameter with good functional results regarding the partial lost of anterior cartilaginous support. Our procedure mimics this technique with the advantage of non compromise the anterior support of the airway. To use pericardium in this case is particularly attractive because the good prognosis and long life expectancy. Long term results in ten of 12 children with APT reported by Ko Bando show absence of stenosis, a low rate of new corrective procedures, and normal growth of repaired tracheas [5]. Others APT reports support these results [6]. We expected that the recipient respiratory epithelium covered the new surface as has been shown by Cheng [7], improving the transport of secretions and the clearance of bacteria and particles. Our patient has not had productive cough and recurrent bacterial bronchitis, suggesting a better clearance of secretions and defense than patients with prosthetic material [8].

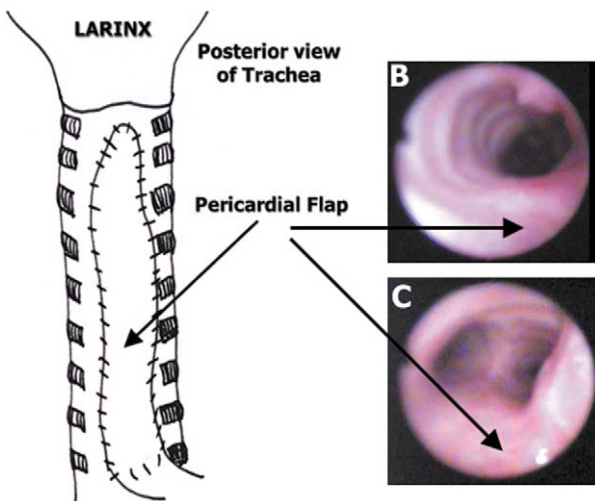


Fig. 2. Scheme of pericardial replacement of membranous wall (left). Flexible bronchoscopy after 5th month. Views of middle and lower third of trachea (B,C).

The indication of stenting in bulge tumors like this is reserved as temporal support of ventilation, while a definitive treatment can be performed. The selection of dynamic stent instead of a tube stent was due to the proximity of the carina with the tumor. However, the flexible design of posterior wall of dynamic stent caused obstruction by posterior tumor protrusion. Stenting for non operable benign tracheal stenosis is controversial, but it can be the unique solution available. Infection, displacement, migration and material fatigue have been reported and the long-term evolution is uncertain, especially when the stent need to be withdrawn [8,9,10].

We think the pericardium is a very good alternative to replace or reinforce MPT, life in trauma when the use of prosthesis or extensive resections are not appropriate.

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