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Cervical esophageal duplication cyst: case report and review of the literature☆

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Abstract Cervical esophageal duplication cysts are rare congenital anomalies that can be successfully managed surgically. These anomalies are rare causes of upper airway obstruction. We present here a case of a cervical esophageal duplication cyst in an infant, along with a review of the literature concerning this anomaly.

Esophageal duplication cysts are benign, congenital, cystic masses that occur in the neck, mediastinum, or both. Isolated duplication cysts of the cervical esophagus are quite rare. These lesions can compromise a normal airway resulting in symptomatic upper airway obstruction. We report an interesting case of an isolated cervical esophageal duplication cyst in an infant and review the available literature on this rare anomaly.

1. Case presentation

A 9.5-month-old healthy boy presented with a 6-month history of biphasic and constant stridor. No prior diagnostic investigations had been undertaken. Complete head and neck examination, including flexible nasopharyngolaryngoscopy, was unremarkable. Radiographic imaging showed an extrinsic mass compressing the trachea at the subglottic level (Fig. 1). Bronchoscopy demonstrated mild tracheomalacia as well as an extrinsic mass extending from the cricoid to the carina and compressing the trachea. Computed tomography (CT) confirmed a large well-circumscribed cystic mass occupying the superior mediastinum, compressing and displacing the trachea (Fig. 2A-C).

Because of the presence of an intrathoracic component, a right thoracoscopic resection was attempted. During thoracoscopy, the cyst was difficult to visualize, partially because of the patient's intolerance of the iatrogenic pneumothorax and right lung collapse. The procedure was converted to an open muscle-sparing right thoracotomy. The distal aspect of the cyst was located just past the

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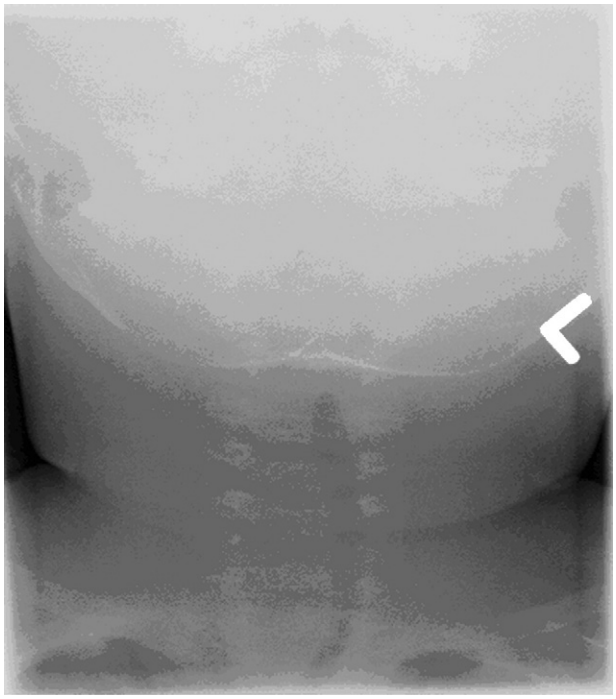

Fig. 1 Anteroposterior view showing extrinsic compression and displacement of the cervical trachea.

thoracic inlet, rendering dissection quite challenging. With traction on the cyst, complete excision was achieved without a cervical incision. However, an esophageal perforation occurred at the site of the common wall of the cyst and esophagus. This was repaired primarily and drained with a chest tube. Postoperatively, the patient was placed on nothing per os. An esophageal contrast study on the sixth postoperative day showed an intact esophagus without a leak. Feeding was gradually advanced, and the patient was discharged on the 10th postoperative day. The patient's stridor gradually improved. At the 18th-month follow-up visit, the child was asymptomatic with no further airway difficulties.

Pathologic examination demonstrated a round unilocular cystic mass (Fig. 3A, B), measuring 2.8 × 2.4 × 1.2 cm. The cystic wall measured 0.2 to 0.3 cm in thickness. The wall was composed of an outer layer of skeletal muscle (Fig. 3C), an inner layer of smooth muscle (Fig. 3D) with some smooth and skeletal muscle intermixed and lined by pseudostratified ciliated respiratory mucosa with occasional submucosal glands (Fig. 3D). The presence of skeletal muscle confirmed the cervical location of the esophageal duplication.

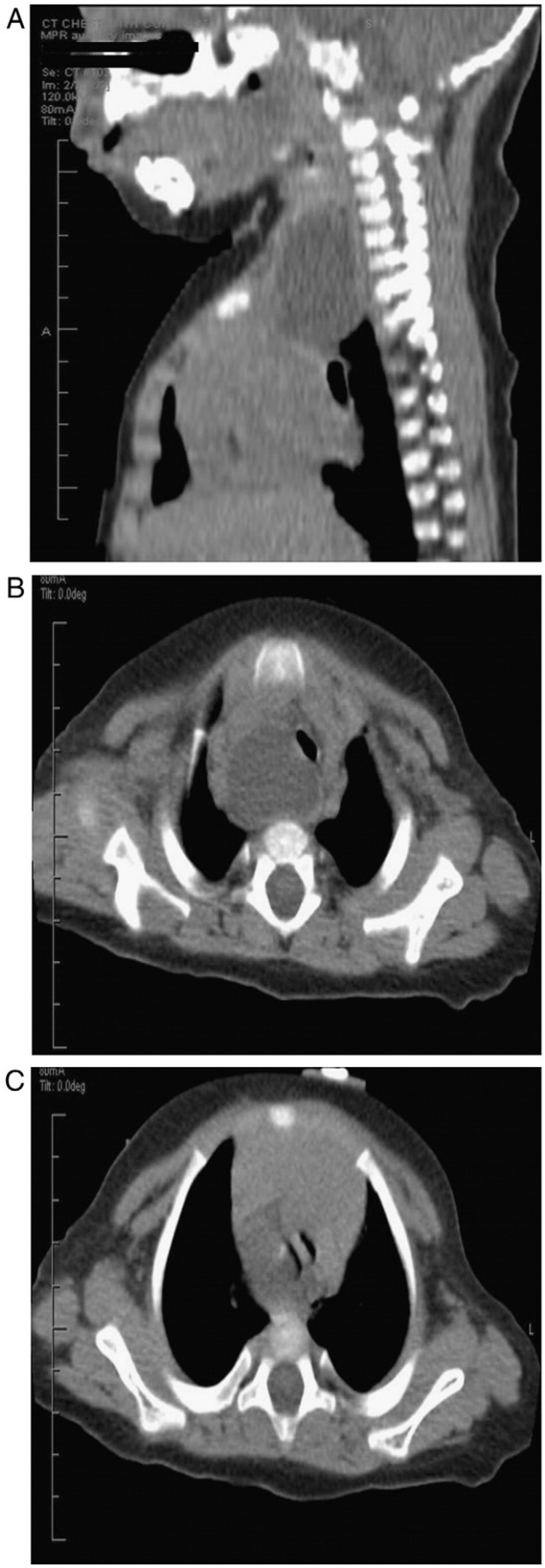

Fig. 2 Sagittal (A) and transverse (B, C) views of the CT scan are shown. The cyst extends past the thoracic inlet on the sagittal view, although its bulk is in the neck. The transverse views show displacement of the thoracic trachea (B) and compression of the carina (C). Despite the findings on CT imaging, the cyst was quite difficult to approach through the mediastinum.