CASE REPORT

The Dilemma of a Lateral Pharyngeal Mass Causing Stridor in a Neonate

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Summary

Neonatal stridor resulting from intrinsic or extrinsic aberration in the upper respiratory tract often poses not only a diagnostic problem, but also a difficult airway and a dilemma as to the necessity / timing of surgical intervention. A 45 day old female child with increasing stridor since birth was managed by emergency intubation and CT scan followed by excision biopsy of the cystic left sided parapharyngeal mass via a transcervical approach. On histopathology, the excised specimen was reported as cystic salivary choristoma.

Key Words: Choristoma, Salivary, Parapharyngeal space, Transcervical

Introduction

Early management of mass effect causing stridor in a neonate is vital as the smaller airway, insufficiently developed cough reflex and easy compressibility makes airway management difficult even for experienced anaesthetists and otolaryngologists. Lesions involving the nasopharynx, pharynx, larynx or upper trachea can cause stridor in a neonate. Parapharyngeal space lesions are an extremely rare cause for neonatal stridor with only a few cases reported in literature.

Choristoma is a related form of heterotopia where one or more mature differentiated tissues aggregate as a tumour-like mass at an inappropriate site. Heterotopic thyroid tissue, ectopic thymus[1] and choristomas of neuroglial[2] and gastric[3] origin in the region of the pharynx can present with stridor in a neonate. In the parapharyngeal space, only choristomas of neuroglial origin have been reported in literature as a cause of airway obstruction[2].

Case Report

In June 2001, a forty-five day old female infant was referred to the ENT Department, Kasturba Medical College Hospital, Manipal, India by the Paediatrician with history of noisy breathing and cyanosis during feeds since birth. On examination, the infant had a respiratory rate of 40 per minute, heart rate of 140 per minute and had suprasternal retraction. The oxygen saturation was 98-99% with cycles of airway obstruction lasting for 3-4 seconds during which the SpO_2 dropped to 86-88%. Plain X-ray soft tissue neck lateral view showed prevertebral widening from C4 to C7 and displacement of the trachea but no tracheal narrowing. Due to the increasing airway obstruction, an emergency awake intubation was done with a 3.5mm endotracheal tube in the computed tomogram scan room. The CT scan done with contrast showed a non enhancing well defined 2.6 by 2.3cm cystic mass in the left parapharyngeal space prestyloid compartment, displacing the fat pad posterolaterally (Fig.1). It was medial to the angle of the left mandible extending to the infratemporal fossa; superiorly, there was no

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evidence of skull base erosion and inferiorly, it extended to just above the level of the hyoid bone. The airway was compromised and displaced to the right but there was no displacement of the internal jugular vein and internal carotid artery. An excision of the cyst under general anaesthesia via a transcervical approach was performed. Using a left curvilinear incision at the level of the hyoid bone, the subplatysmal flap was elevated. The mass was exposed (Fig.2) and removed in toto. Intraoperatively, there was a small tear in the pharyngeal mucosa which was repaired with 4-0 vicryl.

Nasogastric tube was inserted and the child was shifted to the neonatal intensive care unit post operatively. Extubation was performed the following morning. There was a left lower motor neuron facial nerve paresis which probably developed due to traction of the skin flap. Three days later a pharyngocutaneous fistula developed. Both of these were successfully treated by conservative management.

Cut section of the mass revealed a cyst filled with a thick jelly like substance and irregular walls. On histopathological examination, the cyst wall was lined by pseudostratified columnar epithelium with focal areas of metaplastic squamous epithelium. This was overlying closely packed congested vascular channels lined by flattened endothelium, skeletal muscle fibres in which lobules of immature mucinous salivary gland tissue were embedded along with few dilated ducts with outpouchings (Fig.3). Based on these findings, cystic choristoma of salivary origin was reported.

On the last follow up, one and half years post surgery, the child was asymptomatic, thriving well and the scar was only faintly visible.

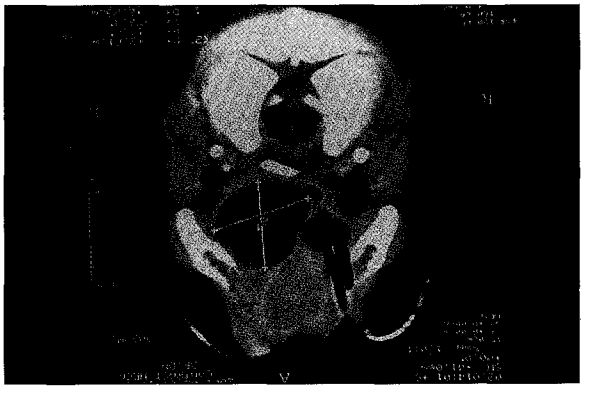

Fig. 1: CT scan (axial cut) - large well defined, non-enhancing cystic mass in the left parapharyngeal space (prestyloid compartment).

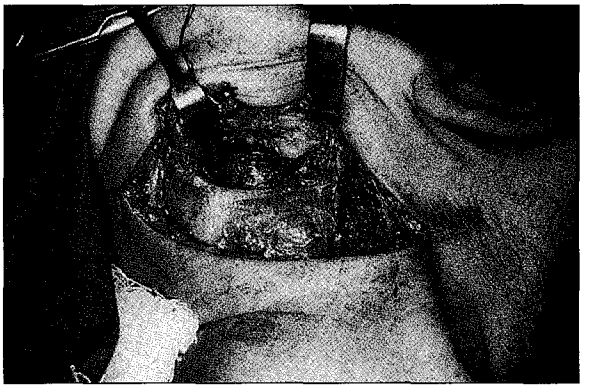

Fig. 2: Left transcervical approach exposing the mass.

Fig. 3: Histopathology (H&E x20).

Discussion

Stridor in a neonate may be caused by lesions in the pharynx, larynx or trachea which may be inflammatory or neoplastic in origin. Non neoplastic lesions in the region of oropharynx reported as a cause of stridor in neonates are hairy polyp[4], lingual thyroid, branchial cyst and thyroglossal duct cyst. Heterotopic neuroglial tissue has been described in the parapharyngeal space presenting with stridor in newborn[2].

Control of the airway in an infant with airway obstruction requires expert anaesthetic intervention. Only following stabilisation of the airway can further investigations like computerised tomographic scan be performed. Though in some cases computerised tomography can be of assistance in airway management as in the above case report.

Management of anticipated airway obstruction by a cystic mass has been attempted by antenatal aspiration with limited success. Following delivery, postnatal aspiration of the cyst prior to clamping of the cord will help avoid a tracheostomy[5].

Choristoma arising from the posterior wall of the laryngopharynx and causing stridor has been successfully treated with CO2 laser[3]. Laser resection is feasible via an endoscopic approach in lesions that arise in the lumen of the oropharynx, larynx or hypopharynx.

Parapharyngeal lesions causing stridor have been removed via a transcervical approach. A high post operative morbidity i.e. lower cranial nerve palsies was reported in cases of neuroglial choristoma[2]. This may be due to fact that neuroglial choristomas are usually not encapsulated and tend to blend or attach to the surrounding structures leading to possible neural injury during resection. Chance of injury to neurovascular structures in the parapharyngeal space is relatively less when the mass is well encapsulated, as in the above case report. Prestyloid compartment lesions have less post-operative complications irrespective of the approach. Facial nerve palsy can be avoided by tracing the nerve. Salivary fistulae can occur, but it usually heals with conservative management. The region of the pharyngeal wall that is thinned out due to tumour expansion can be sloughed out leading to a pharyngeal fistula as found in our case. This can be prevented by resecting the thinned out portion of the mucosa and closing primarily.

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