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Laryngeal cysts in infants and children—A pathologist's perspective (with review of literature)

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ABSTRACT

Objectives: To expose the rarity of the phenomena of congenital laryngeal cysts. Additionally, a discussion is presented in support of the basic similarities between laryngeal duplication cyst and bronchogenic cyst and a proposal to use the term 'bronchogenic cyst' (appended by the location) is put forth.

Materials and methods: Three cases of cysts of the larynx, two in the neonate and one, in a child were discovered in the pediatric age group from the archives of the Department of Pathology, Kasturba Medical College, Mangalore.

Observations: Laryngeal duplication cyst/bronchogenic cyst in a 6-day-old male infant was lined by predominantly respiratory with focal squamous epithelium. The wall contained fibromuscular bundles within a myxoid stroma studded with seromucinous glands. The fibromuscular bundles were positive for smooth muscle actin (SMA) and negative with desmin. A 3-year-old male child diagnosed of laryngocele had the cyst lined by respiratory epithelium and the wall densely infiltrated with lymphoid cells. A 20-day-old male neonate with laryngomalacia had a small arytenoid cyst lined by squamous epithelium.

Conclusion: On morphological grounds, a laryngeal duplication cyst may be called a bronchogenic cyst and published reports do not deny that both these entities are cognate. Hence, pathologists should enjoy the freedom to write 'bronchogenic cyst' as the final impression if the histological features should so indicate. Otolaryngologtists however might term the congenital cyst as Type 1, 2a or 2b (according to Forte's classification) taking into account the laryngoscopic, radiologic, intraoperative and pathological findings.

1. Introduction

Congenital cysts in larynx, though rare phenomena are tangibly precarious to the newborn or the infant since they frequently cause stridor, thence to airway blockage eventually resulting in many instances to severe morbidity, and in some, death [1–3]. Predominantly these cysts are located in the supraglottis or glottis proper, the subglottis being the minimally favored site [4]. Parenthetically, subglottic cysts more often result from complication of intubated preterm infants, perhaps due to inflammation and secondary cyst formation consequent on obtruding upon the salivary duct [1]. A few of the congenital cysts distort the cartilages of larynx, attach, dent or prolapse through the laryngeal wall and may sometimes exhibit an intralaryngeal swelling with an extralaryngeal annex causing their excision possible only by open surgery [2,3]. The composition of the cyst wall often can be traced embryologically to contain derivatives of the endoderm and the mesoderm implying that they are of the ilk of bronchogenic cysts or more primitively, bronchopulmonary foregut malformations. In these cases, though the otorhinolaryngeal surgeons prefer to term them 'laryngeal duplication cyst', the pathologists remain at a loss to comprehend the difference between this entity and the histology of another widely accepted lesion known as the 'bronchogenic cyst'. In the current study, the department records of 6 and half years were reviewed, from which 3 laryngeal cysts in the pediatric age group were uncovered.

2. Materials and methods

The laryngeal cystic lesions in the pediatric age group were collected from the archives of the Department of Pathology in Kasturba Medical College, Mangalore of Manipal University, India. From January 2006 till June 2012, three cysts in the larynx of children were identified. The medical record of case 1 was studied

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in complete detail. The other two were referral cases and their details obtained from the histopathological requisition forms were sketchy.

3. Observations

3.1. Case 1

A 6-day-old full term male neonate delivered vaginally was referred for ENT consultation for stridor. His antenatal history was associated with polyhydramnios. His perinatal period was complicated by respiratory distress starting 6 h after birth to relieve which, he was intubated. Attempts at extubation were met with recurring failures as the repiratory distress remained relentless. Hemogram was well within normal limits for his age. Suction from endotracheal tube sent for cytology showed sheets of neutrophils. Culture sensitivity revealed gram negative bacilli along with staphylococcus.

An initial computed tomogram (CT scan) (Fig. 1A) conveyed an unsure opinion of esophageal duplication cyst based upon the observation of a well defined non-enhancing hypodense lesion in the left paratracheal region (1.6 cm × 1.2 cm × 1.1 cm) displacing the trachea towards the right and compressing the proximal 1/3rd of esophagus. A direct laryngoscopy exposed a large thin walled cyst distending the left aryepiglottic fold and false cord. The glottic chink was not visible. A repeat CT scan, impelled by the laryngoscopic discovery revealed the extent of the cystic lesion seen in left paralaryngeal region from just below the hyoid superiorly up to the superior pole of left lobe of thyroid inferiorly (Fig. 1B). Its measurement was unchanged. The lesion compressed the apex of the left pyriform sinus and displaced the trachea to the right. An impression of laryngeal cyst was imparted.

The cyst was excised by exploring the thyrohyoid space through a subhyoid incision under general anesthesia. The tense cyst, stretching and thinning out the laryngeal mucosa arose between the thyroid ala, the laryngeal and pharyngeal mucosa. The complete intralaryngeal breach was filled with sticky material.

The postoperative period was uneventful with 3 days of endotracheal tube in situ followed by extubation with minimal stridor. Fibreoptic bronchoscopy showed edematous aryepiglottic folds for which systemic steroids were given. The patient was discharged on the 8th postoperative day and was doing well at 6 months follow up.

Microscopically the cyst was mostly lined by respiratory (Fig. 2A) and focally by non-keratinizing squamous epithelium (Fig. 2B). Immediately beneath the epithelial lining, a thin fibrous bundle was seen surrounded by a myxoid stroma in which seromucinous glands and maturing cartilage were embedded (Fig. 2A). There were no misgivings about this archetypal description of bronchogenic cyst. Since the term 'bronchogenic cyst' was construed as directly contradicting the clinical diagnosis 'laryngeal duplication cyst' as proposed by otolaryngologists, immunohistochemistry with smooth muscle actin (SMA) and desmin was performed to prove conclusively the presence or absence of smooth muscle in the wall of the cyst. Though desmin was negative (Fig. 3B), SMA was positive (Fig. 3A) in the bundle of spindled cells encasing the epithelium.

3.2. Case 2

Mild hoarseness of voice and difficulty in swallowing were the introductory symptoms in a 3-year-old male child. With normal vital signs and preliminary laboratory reports that included a complete hemogram, the pediatrician was unable to ascertain the source of the symptoms and thus referred him to the otorhinolaryngology department. An indistinct translucent fullness over the left vestibular fold was noted on direct laryngoscopy. CT scan exposed a cyst in the left false cord. Diagnostic aspiration did not yield any fluid. Since the symptoms were progressive, a decision to excise the cyst by lateral cervical approach to the thyrohyoid membrane was made and the excised cyst was sent for histopathology. Four pale white membranous tissue fragments together measuring 0.3 cm across were received. The microscopy was described as thin fibrous tissue infiltrated heavily with lymphoid cells lined by respiratory epithelium. The final verdict confirmed the clinical suspicion of laryngocele.

3.3. Case 3

An ENT opinion was immediately asked for this neonate who was beset by respiratory distress at birth. Immediate intubation relieved his cyanosis but severe stridor ensued after the endotracheal tube was removed to enable the ENT surgeons to view the larynx. Flexible bronchoscopy displayed a small, approximately 0.5 cm cystic lesion towards the lateral aspect of right arytenoid. The lateral aryepiglottic folds were seen obtruding into the laryngeal inlet. They were also mildly swollen and the

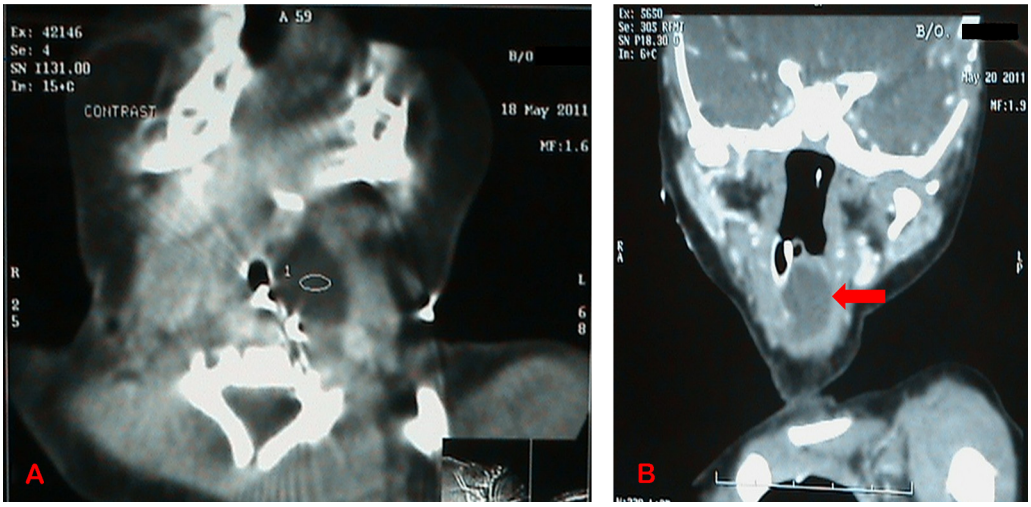

Fig. 1. (A) CT scan showing well defined non-enhancing hypodense lesion in the left paratracheal region. (B) CT scan showing the cystic lesion in the left paralaryngeal region. CT scan was done after laryngoscopy.