Addressing a rare cause of paediatric stridor: subglottic cyst

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DESCRIPTION

An 18-month-old girl child was brought by her parents to emergency service with noisy breathing and increased breathing effort progressive over several months, but significantly worsened for 1 week following fever and diarrhoea. She had a weak cry with no difficulty in feeding and coughing or choking episodes. She was hospitalised and received treatment for croup multiple times in the past without complete symptomatic relief. The child was born preterm and kept intubated for a month after birth. At presentation, child was irritable with tachypnoea and tachycardia without cyanosis. Her biphasic stridor with suprasternal and intercostal retraction worsened on crying and was not relieved on postural changes. Due to her symptomatology during the ongoing COVID-19 pandemic, the COVID-19 test was sent and endoscopy was avoided. Significant subglottic airway compromise was noted in the lateral neck radiograph (figure 1A). The contrast-enhanced CT of neck revealed a hypodense, non-enhancing lesion measuring 9×8 mm in the subglottis (figure 1B). With a presumptive diagnosis of subglottic cyst and a negative COVID-19 test, the child was taken to operating room and tracheotomy was performed with mask ventilation. Direct laryngoscopy revealed a submucosal cystic lesion immediately beneath the mobile vocal cords without any significant patent airway around the cyst (figure 2A). Using a 22-gauge microlaryngeal needle, 1 mL of thick yellowish clear fluid was aspirated from the cyst. The cyst arising from the right posterolateral wall of subglottis was marsupialised using coblator. Through the adequately patent airway tracheoscopy was performed which was unremarkable (figure 2B). Postoperatively, child recovered well and was discharged with a corked tracheostomy tube and nasal breathing. Follow-up endoscopic evaluation at 2 months did not suggest cyst

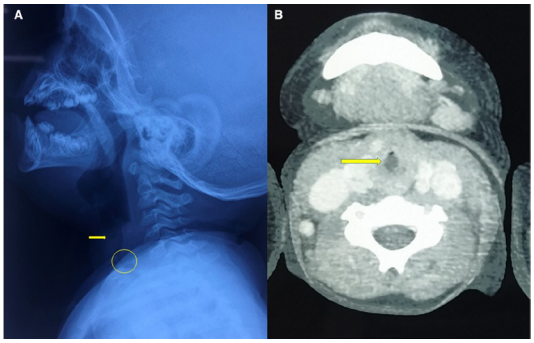

Figure 1 Preoperative neck soft tissue radiogram (lateral view) showing a lesion in the subglottis with significant airway narrowing (A, yellow pointer shows the laryngeal ventricle and subglottic cyst compromising airway shown in yellow circle) and contrast-enhanced CT (axial view) showing a hypodense, non-enhancing lesion in the subglottis with only minimally patent airway anterior to it (B).

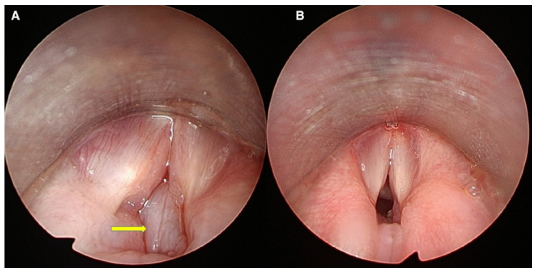

Figure 2 Direct laryngoscopy image showing mucosa covered cyst (arrow) beneath the true vocal cords (A) and patent subglottic airway following marsupialisation of the cyst (B).

Patient's parent perspective

Our child's breathing was noisy and cry was weak since the day she was born. She had received treatment many a times in different hospitals and she had some temporary relief each time. However, her breathing had worsened over many months and we were really frightened with her loud breathing while she cried and while she played for last 1 week. Recently, after the CT scan, we were informed about a fluid filled sac in her windpipe which was growing slowly and causing her struggle to breathe. It was also very difficult for us to visit hospitals in this difficult period due to the coronavirus infection and lock down. Doctors had informed us about the need for temporary tracheotomy and how to take care of it. Now, after the operation, our child is breathing comfortably while playing even with closed tracheostomy tube and there is no problem with her feeding. We are able to take care of her tracheostomy tube at home as instructed by the doctors and the tube will be removed after repeat check-up in the hospital. Doctors have counselled us for the need for repeated follow-up so as not to miss the disease if it comes back in which case another surgery might be needed. We are very hopeful as the doctors have assured that our daughter will have comfortable breathing, feeding and good voice after the tube is removed.

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Learning points

- Prematurity has been associated with congenital subglottic cyst and history of intubation has also been described in few cases suggesting a possible acquired aetiology.
- Direct laryngoscopy is the gold standard for making a diagnosis of this uncommon cause of paediatric stridor.
- Rigid endoscopic instrumentation to rupture the cyst or needle aspiration can lead to immediate symptomatic relief but should be followed by definite endoscopic marsupialisation to avoid recurrence.

recurrence. Delayed decannulation following exclusion of recurrence has been planned to ensure avoidance of any emergent visit to hospital during the ongoing COVID-19 pandemic and nation-wide lockdown.

Subglottic cysts are retention cysts of subepithelial mucosal glands which may be either congenital or acquired. This rare cause of paediatric stridor can be single or multiple and commonly arise in the posterolateral subglottis with circumferential extension or involvement of upper trachea.[1] Preterm infants seem to be at greatest risk for developing subglottic cysts as frequent endotracheal intubation may lead to mucosal trauma, inflammation, duct occlusion and subsequent cyst formation.[2] The clinical importance of subglottic cysts is dependent on the degree of airway compromise, as small and solitary cysts may be an incidental finding on endoscopy and large or multiple cysts may cause catastrophic airway obstruction. Subglottic cysts should be differentiated from hemangioma, stenosis, respiratory papillomatosis which are more common causes of paediatric stridor and rare subglottic lesions such as thyroglossal cysts and ectopic thymic cysts.[3] Diagnosis is suggested by radiology and confirmed by direct endoscopic visualisation under anaesthesia.[4]

Treatment modalities include decompression with rigid instrumentation, excision or marsupialisation using cold instruments.[5] Laser, microdebrider and coblator are used without creating the circumferential raw area and have been associated with lesser recurrence and subglottic scarring.[6] Follow-up endoscopy is recommended to detect recurrence cyst formation which is not unusual.

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