

# External laryngocoele: A rare cause of stridor in neonate

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## ABSTRACT

A laryngocoele is an abnormal dilatation of the laryngeal saccule. It is a rare cause of stridor in the newborn or in early infancy. Most laryngocoeles are asymptomatic but symptoms of hoarseness, feeding difficulty, lump in the neck and upper airway obstruction may occur. We report a case of external laryngocoele causing partial airway obstruction in early infancy. Spontaneous resolution occurred following conservative medical management thus avoiding surgical intervention.

**Keywords:** Laryngocoele, external, infant, complications, respiratory distress

## INTRODUCTION

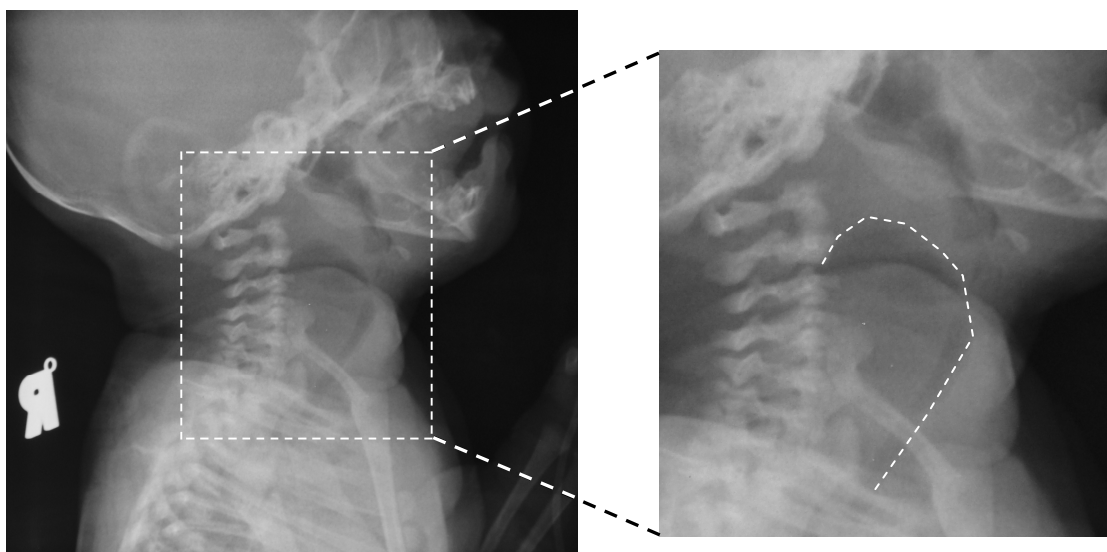
A laryngocoele is an air-filled dilatation of the saccule of the larynx. The pathogenesis of laryngocoele is still unclear. It is a rare condition especially in the paediatric population. In a review of 139 cases of laryngocoele in general, the peak incidence was in the sixth decade with two out of three laryngocoeles being unilateral and the rest being combined, external or internal with roughly equal frequency. <sup>1</sup> If found in an infant, it is invariably congenital in nature. <sup>2</sup> The clinical presentation of laryngocoele in an infant includes respiratory distress, inaudible or hoarse crying, feeding dif-

faculties and neck mass. However, some laryngocoeles may go unnoticed and may be detected accidentally on routine neck radiograph. In the presence of acute airway obstruction, delays in diagnosis can lead to a fatal outcome.

## CASE REPORT

A four-day-old baby girl was admitted to a newborn intensive care unit for a huge left neck swelling causing airway compromise. She was a term baby weighing 2.3kg after a normal pregnancy and had no risk factors for perinatal sepsis. The parents noticed noisy breathing since day one of life which was unrelated to posture. However, it was associated with feeding difficulty and recurrent episodes of regurgitation. A rapidly enlarging

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**Fig 1: A lateral soft tissue neck x-ray showed an air sac in the antero-superior aspect of the larynx highlighted by box. Magnified image of the box showing the air sac of the laryngocoele (outlined with broken white line).**

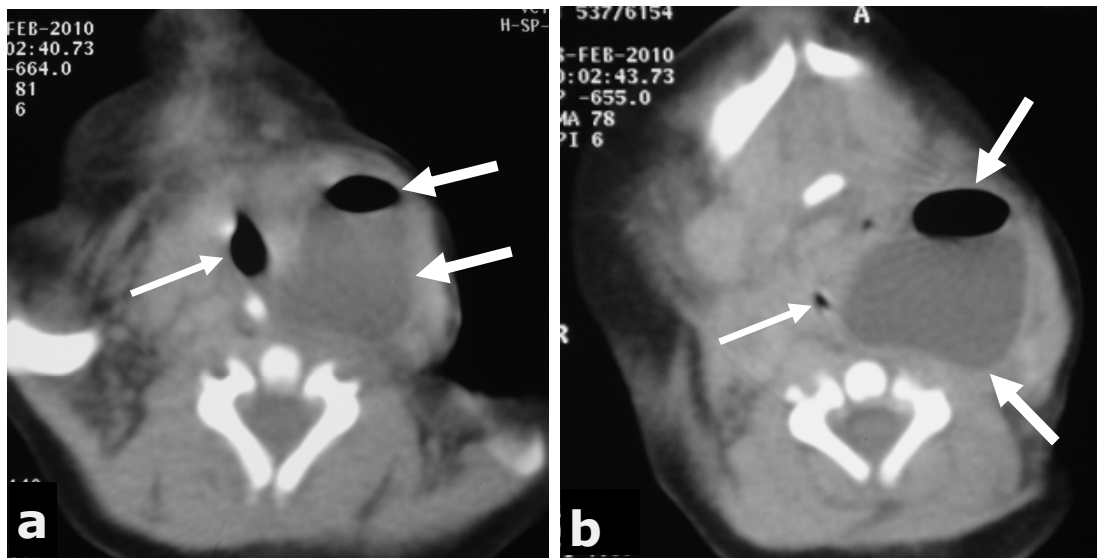
neck swelling appeared on day four of life accompanied by signs of respiratory distress. The condition was accompanied by history of a low grade fever for one day duration.

Physical examination revealed a high pitched inspiratory stridor with deep sternal recession. The patient was febrile. On the left side of the neck, a soft and non-tender mass measuring 3cm in its greatest dimension was palpable. It appeared to increase in size when the patient cried. She was kept under observation and it was decided she was not for intubation as her respiratory distress was not severe. She was put under a head box with oxygen 5L/min and covered with intravenous C-Penicillin 230,000ii b.i.d (100,000 ii/kg/dose) and gentamicin 9 mg daily (4mg/kg/dose) for presumed sepsis following the National Antibiotic Guidelines 2008. Intravenous antibiotic was discontinued after five days as the blood culture was negative. Flexible fibre-optic laryngoscopy examination was normal.

A plain neck x-ray showed a well de-

defined round radiolucent area protruding into the soft tissue of the neck (Figure 1). Computed tomography (CT) scanning with contrast demonstrated a cystic mass with an air-fluid level in the left paralaryngeal space (Figure 2). The lesion caused mass effect to the larynx with obliteration of airway at the C1 level (Figure 3). There was no evidence of intralaryngeal pathology.

After a few days of antibiotic therapy, the stridor regressed and the neck swelling progressively resolved. Repeat assessment at two weeks later revealed that the child was active without any respiratory or feeding problem. There was no visible or palpable mass in the neck. The initial decision to excise the lesion via an external approach was deferred with close follow-up of the patient. At 1 year follow up, the child remains asymptomatic and thriving. The parents were advised to be compliant to follow up as excision of the lesion may be considered if the patient becomes symptomatic in the future.



**Fig 2:** Axial computed tomographic images showing a) external laryngocoele as a cystic fluid filled mass (large arrows) on the left side with the trachea indicated by small arrow and b) laryngocoele causing compression of the airway.

## DISCUSSION

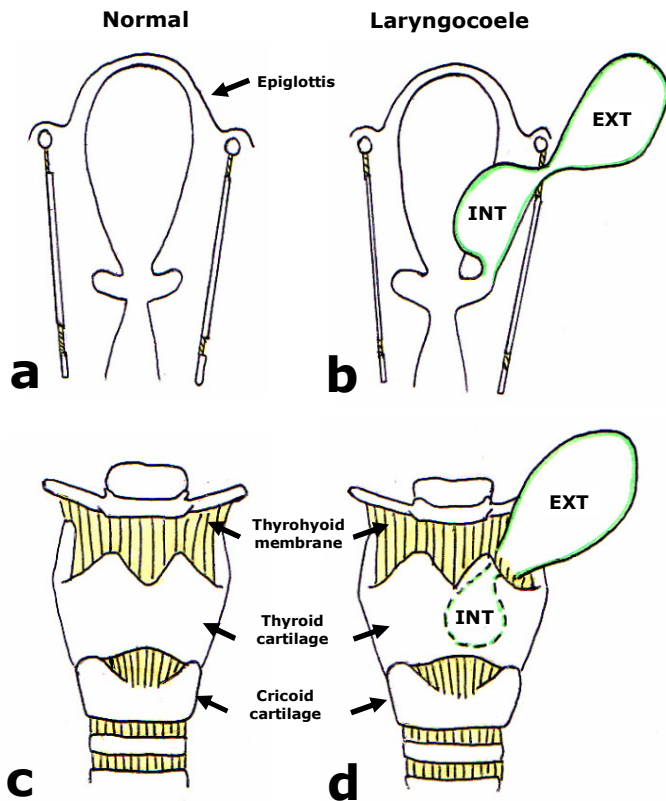
A laryngocoele is an air-filled dilation or herniation of the saccule and it communicates with the lumen of the larynx. It is uncommon in infants, with peak incidence in the sixth decades of life. Even though it is very rare, the diagnosis should be considered as one of the causes of progressive upper airway obstruction in the neonatal period or early infancy.<sup>3</sup>

There are three types of laryngocoele; internal, external and mixed. An internal laryngocoele is confined to the interior of the larynx and extends postero-superiorly into the area of the false vocal cord and aryepiglottic fold. An external laryngocoele extends cephalad to protrude laterally into the neck through the thyrohyoid membrane. When both components present together, it is termed a mixed or combined laryngocoele.<sup>4</sup>

The aetiology of laryngocoeles is unknown. It could be congenital and acquired.<sup>1</sup>

The laryngocoeles observed in newborns are certainly congenital, while in acquired they tend to be associated with activities that increased intralaryngeal pressure like glass blowing and playing wind instruments.<sup>2</sup> These activities require high intralaryngeal pressure to be created repeatedly, and thus explain the mechanism of herniation of the laryngeal sac-cule.

The symptoms depend upon the type of laryngocoele and the severity is partly dependent on the size. Most laryngocoeles are asymptomatic and incidentally discovered during routine neck radiographs. Classically an external laryngocoele presents clinically as a swelling in the neck which will increase during the Valsalva manoeuvre and become smaller on palpation. When a large external laryngocoele produces pressure and displaces the larynx, it can cause an airway obstruction and even death. Internal laryngocoeles manifest with hoarseness, dyspnoea, cough and



**Fig. 3: Illustrations showing normal structures (a and c) compared to laryngocoele (b and d): internal (INT) and external (EXT) laryngocoeles are outlined by green margins, the origin from the laryngeal ventricle (saccule is located at the apex of ventricle) and the extension of external type beyond thyrohyoid membrane. Structures painted in yellow are laryngeal membranes.**

foreign body sensation. Infection of a laryngocoele can cause acute symptoms or worsen existing symptoms.<sup>5</sup>

Our patient presented with early symptoms of stridor and feeding difficulty. The neck swelling only became apparent and associated with worsening of the respiratory symptoms when she had fever. A plain neck radiograph is sufficient to demonstrate the air filled sac protruding from the soft tissues of neck. However, a CT scan is more valuable as it can show a combined laryngocoele when only one component is clinically suspected.<sup>6</sup>

Subclinical congenital laryngocoeles may be managed conservatively provided there is no airway compromise. When symptomatic, external laryngocoeles are preferably removed by an external surgical approach.<sup>7</sup>

In this case, a dramatic improvement of symptoms upon administration of antibiotics may have suggested an infected laryngocoele as a cause of progressive enlargement of the mass leading to airway obstruction.

Although the CT scan demonstrated an air fluid level, there was no thickening of the walls or rim enhancement of the laryngocoele that would suggest a laryngopyocoele needing surgical drainage. It has been reported that eight percent of laryngocoeles get infected and present as laryngopyocoele.<sup>1</sup> Our patient responding well with the conservative management and the neck mass completely subsided. Removal of the lesion may be considered in future if the patient become symptomatic.

In conclusion, our case highlighted a

rare cause of stridor in a infant. Clinicians, including front line clinicians and pediatricians needs to be aware and consider laryngocele as a cause of stridor or upper airway obstruction.

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## Brunei Darussalam – Healthcare in Pictures



The first intake of nurse trainees in Brunei based in the Brunei General Hospital (Picture taken in 1953).

Top row from the left: Helen Chow, Grace Yapp, Sister Maddock, May @ Hjh Rokiah and Susun Yong  
Bottom row from the left: Cek Sum, Rogayah, Datin Marianna Hart (*spouse of Dato Peter Loy de Villiers Hart, refer to Healthcare Pioneers Brunei International Med J 2011; 7 (4):191*) and Datin Hjh Habibah (Matron of Nursing, Ministry of Health Brunei Darussalam).