The Journal of Laryngology and Otology
May 1992, Vol. **106**, pp. 448–449

Neonatal vallecular cysts and failure to thrive

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Abstract
The case of a vallecular cyst in a neonate is described. The presentation was with failure to thrive. This previously unreported mode of presentation is discussed, and aspects of the management are emphasized. In particular, the value of flexible nasopharyngoscopy in assessing the neonatal laryngopharynx is highlighted.

Introduction

Congenital cysts of the larynx are rare. Because of their potential for morbidity and mortality, they have stimulated interest for many years. The earliest English language publication on the subject was by Abercrombie in 1881. The case which we report illustrates a most unusual presentation of a laryngeal cyst—that of failure to thrive. Just as all clinicians treating children should be aware that this common paediatric problem can be due to unusual and potentially serious upper aero-digestive tract pathology, ENT surgeons should be familiar with this possible presentation of a vallecular cyst.

Case report

A 12-day-old Nepali male child was admitted to the Paediatric Department with failure to thrive. He had been born uneventfully at full term following a normal pregnancy, and there were no perinatal problems. His birthweight was 3.1 kg and Apgar scores were normal. The baby was breast-fed, but failed to regain his birthweight. No abnormality relating to breathing, crying or feeding had been noted until at 12 days a short episode of slight stridor associated with mild cyanosis during a feed precipitated admission. Examination at this time revealed a well baby with no abnormal physical signs, but failure to regain birthweight was confirmed. There was no stridor or intercostal recession, a normal sounding cry was noted, and chest examination was unremarkable. There was no change with variation in posture. All laboratory investigations and the chest X-ray were normal.

Five days after admission, aged 17 days, the baby was noted to be cyanosed around the mouth after a feed. In the following 24 hours three similar episodes of cyanosis after feeds were noted and ENT advice was sought.

At ENT assessment the child appeared well, although underweight. An Olympus ENF P 3.6 mm OD flexible nasopharyngoscope was passed transnasally into the pharynx. A large smooth cystic swelling was seen arising from the left vallecula obscuring the left side of the laryngeal inlet. There was no airway impairment, and no other abnormality was noted.

At direct laryngoscopy a 1.5 cm diameter broadly based cyst was found arising from the left vallecula attached to the left lateral pharyngeal wall and the left side of the lingual aspect of the epiglottis. The cyst wall was removed with sharp dissection after 2.5 ml of milky fluid content had first been aspirated.

As much of the lining of the cyst as possible was removed by eversion and dissection. The child made an uneventful recovery and fed normally once awake. A repeat flexible nasopharyngoscopy on the fourth post-operative day showed a normal valecula, epiglottis and supraglottic airway, with a small area of exudate over the excision site. There were no further airway problems.

He left hospital six days post-operatively at age 29 days. Figure 1 shows his weight from birth, in relation to the 10th, 50th and 90th centiles. The delay in weight gain until surgery is clear. At review six months after surgery he was in perfect health.

Histology of the cyst lining showed the presence of fibrous tissue with cystic spaces lined by stratified squamous epithelium.

Discussion

Laryngeal cysts in children are rare, the first reported case having been published as early as 1881 by Abercrombie. In 1987, Mitchell *et al.* published the largest single series consisting of 20 cases reflecting the experience of the Hospital for Sick Children, London over a 15-year period. In 1961, Birch reported his analysis of 200 unselected infants and children presenting with stridor to the same institution. Four of these (two per cent) had laryngeal cysts. In 1980, Holinger reported on 219 children presenting with stridor to two Chicago paediatric units over four years. Two of these cases (0.9 per cent) were due to laryngeal cysts.

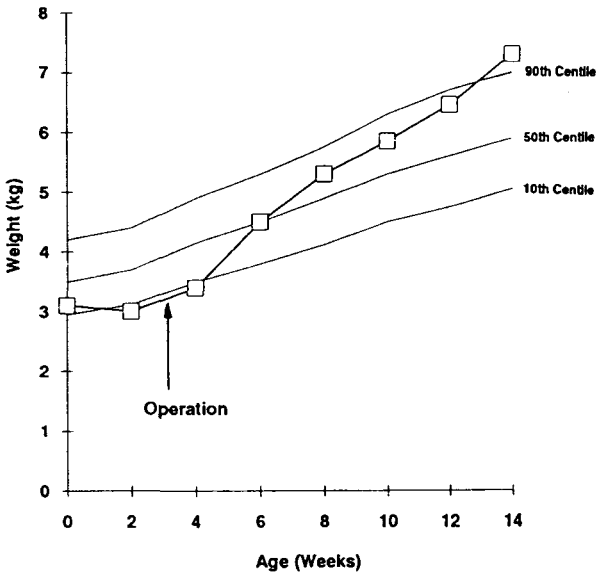

FIG. 1
Serial weights of the child in relation to 10th, 50th and 90th centiles.

Accepted for publication: 30 December 1991.

The De Santo classification of laryngeal cysts into two types—saccular and ductal—is widely accepted (De Santo *et al.*, 1970). Ductal cysts, otherwise known as mucous or retention cysts are more frequently encountered, and are thought to originate from obstructed submucosal glands. They occur anywhere in the larynx where glands are present, but are found most frequently in the vallecula. The cyst found in the child reported here appears to be of the ductal type. Saccular cysts are encountered less commonly and occur in the plane of the saccule. They are thought to result from cystic distention of this structure. They differ from laryngocoeles only in that they contain mucus, while laryngocoeles contain air.

Stridor is the clinical presentation of congenital laryngeal cysts most frequently described. Less frequently, the presentation may involve dyspnoea, an abnormal cry, coughing and episodes of cyanosis. The child reported here presented primarily with failure to thrive. After birth there were no features of upper airway obstruction and nothing to indicate feeding difficulties apart from the observation that feed volumes were small. It was only at 12 days that a feed was complicated by some 'noisy breathing' and slight circumoral cyanosis. Recurrence of a similar episode five days later focussed attention on an upper aero-digestive tract abnormality as the likely aetiology and appropriate referral was made.

The characteristics which determine the mode of presentation of laryngeal cysts appear to be their position and size. In reporting their Mayo Clinic series, De Santo *et al.* (1970) found that 52 per cent of 238 laryngeal cysts studied originated from the epiglottis. The most common sites were its lingual surface, the aryepiglottic fold and then the ventricle and piriform sinus. The findings of Mitchell *et al.* (1987) are similar. A small vallecular cyst is less likely to present with stridor than is an aryepiglottic fold cyst because the latter is so much closer to obstructing the airway itself. The small vallecular cyst is also likely to cause little impairment to swallowing. However, as mucus production distends it further, the cyst will progressively interfere with swallowing. A stage will be reached when the airway becomes compromised. This sequence of events would appear to explain the clinical course of the child in this report. There was initially no respiratory difficulty nor obvious swallowing problem. Small feed volumes were then noted, and the failure to gain weight became apparent. There was still no obvious difficulty in swallowing. The first episode of mild stridor and cyanosis during a feed occurred only on the 12th day. This progressive interference with upper aerodigestive tract function correlates well with the postulated expansion of the vallecular cyst, which reached the considerable size of 1.5 cm in diameter. By contrast, a cyst impinging more directly on the airway, as in the aryepiglottic fold, is likely to cause earlier symptoms of airway obstruction (Birch, 1961; Abramson and Zielinski, 1984; Kristensen and Tveteras, 1986; Mitchell *et al.*, 1987; Bais *et al.*, 1989).

Established or incipient airway obstruction is the most important complication of laryngeal cysts. This can be rapidly accelerated by upper respiratory infection. It is noteworthy that a siginficant number of these cysts are confirmed only at autopsy (Suehs and Powell, 1967). Thorough upper airway assessment and early accurate diagnosis in all likely cases are thus essential.

Most authorities advocate a policy of early direct laryngoscopy under general anaesthetic for all neonates and infants in whom an upper aero-digestive tract abnormality is suspected (Abramson and Zielinski, 1984; Henderson *et al.*, 1985; Kristensen and Tveteras, 1986; Bais *et al.*, 1989), This facilitates a full assessment of the upper airway early and confirmation of the diagnosis. Potentially dangerous delay in establishing the correct diagnosis can otherwise occur and must be avoided (Hollinger, 1980).

As illustrated by its use in this case, we find transnasal flexible nasopharyngoscopy invaluable. The procedure is quick and straightforward to perform, requiring no anaesthetic, premedication or special facilities other than the endoscope itself and a secure hold on the child. This procedure is undertaken at the first assessment and in most cases a diagnosis and treatment plan are established quickly. Where a diagnosis is made that does not indicate an early general anaesthetic procedure, progress can be monitored with repeated flexible endoscopic examinations. This practice is supported by other authors (Vaughty and Reddy, 1980; Nussbaum, 1983). The former records the effective use of this technique on a one day old neonate without difficulty (Nussbaum, 1983). Ideally the cyst lining should be removed completely in order to minimize the chance of recurrence. Mere drainage without any excision of the cyst lining is likely to result in recurrence (Abramson and Zielinski, 1984; Henderson *et al.*, 1985; Kristensen and Tveteras, 1986; Mitchell *et al.*, 1987). Aspirating the contents of the cyst with a wide-bore needle and syringe before removing it is helpful, and is accepted practice (Kristensen and Tveteras, 1986; Mitchell *et al.*, 1987; Bais *et al.*, 1989).

The cyst is then dissected free, using a technique of partial eversion and sharp dissection. Suction is applied to the cyst lining, the negative pressure achieving the necessary eversion. The carbon dioxide laser is reported to be helpful for this dissection, or to vaporize the cyst lining (Abramson and Zielinski, 1984). This technique is awkward in a neonate due to limited access and difficulty in stabilizing the laryngoscope.

In cases of very large cysts, laryngofissure may be required.

Conclusion

Failure to thrive has not hitherto been reported as a presentation of a congenital laryngeal cyst. We emphasize the usefulness of the flexible nasopharyngoscope in establishing an early diagnosis in all patients where an upper aero-digestive tract problem is suspected, particularly in very small babies. The great importance of early diagnosis and treatment of congenital laryngeal cysts is highlighted in view of their potentially severe morbidity and mortality.

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Key words: Failure to thrive; Infant, newborn; Cyst, vallecular