Case report

Failed videolaryngoscopy in an infant with giant vallecular cyst managed successfully with a 'three-person technique' of fibre-optic intubation

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SUMMARY

Vallecular cysts are a rare entity but can complicate the airway management due to the physical impediment and distortion of the laryngeal inlet. It can lead to fatal life-threatening airway obstruction postinduction of anaesthesia. Infancy compounds the risks due to lack of cooperation and physiological reserves. The literature review suggested most anaesthesiologists avoided muscle relaxants and aspiration of the cyst was used as a rescue technique. The present case report illustrates the successful airway management of an infant with a huge vallecular cyst using fibre-optic intubation by a 'three-person' technique after paraglossal videolaryngoscopy failed to secure the airway.

BACKGROUND

Vallecular cysts are a rare cause of upper airway obstruction and can occur due to congenital malformation or acquired ductal obstruction of mucous glands.¹ It usually presents as inspiratory stridor, dyspnoea, dysphagia, abnormal cry, poor feeding, cyanotic and choking episodes, failure to thrive and apnoea. It may also lead to supraglottic airway obstruction due to its mass effect in the oropharynx and posteroinferior sagging of the epiglottis over the glottic aperture.^{1,2} It has been associated with grave consequences such as respiratory failure, need for tracheostomy and even death in past reports.¹⁻³ Airway management of such cases has not been standardised due to the infrequent reporting and depends on the anaesthesiologist discretion. There have been reports of difficult direct laryngoscopy, fibre-optic bronchoscopy (FOB) and even tracheostomy for airway management in such cases.³⁻⁶ Videolaryngoscopy (VL) is an upcoming technology in paediatric airway management and can be an option in such cases. We are reporting this a case of vallecular cyst for its rarity, to emphasise that VL is not the solution in every difficult airway scenario and that 'three-person technique' can be useful for fibre-optic intubation in patients with supraglottic mass lesions. We have also reviewed the airway management of infants with vallecular cyst reported in the literature.

CASE PRESENTATION

A 1-year-old male child, weighing 5.3 kg, presented with progressive difficulty in swallowing solids, choking episodes while feeding, hoarse cry, difficulty

lying supine, failure to thrive, noisy breathing and respiratory distress on feeding. He was being fed only small amounts of liquids and was referred to our hospital from a peripheral centre because of anticipated difficult intubation. On examination, the infant was febrile, irritable, dehydrated, tachypnoeic and had chest wall retraction. A large cystic mass (2×2 cm) was observed occupying the right side of the base of the tongue and crossing the midline (figure 1).

INVESTIGATIONS

A CT scan revealed a large hypodense cystic swelling at the base of the tongue and anterior pharyngeal wall. Laboratory investigations were unremarkable.

TREATMENT

After initial stabilisation, the patient was posted for complete excision of the cyst under general anaesthesia (GA). The child's parents were counselled regarding the nature of difficult airway and informed consent was taken. We planned to intubate the child under inhalational induction with preserved spontaneous respiration. The ENT surgeons were also asked to be prepared for emergency tracheostomy. The airway was topicalised by nebulisation using 2 mL of 1% lignocaine. One drop of 0.05% xylometazoline was instilled in each nostril. In the operation theatre, standard monitors were attached and injection glycopyrrolate 0.05 mg, hydrocortisone 25 mg and dexamethasone 0.5 mg were administered intravenously. After preoxygenation with 100% oxygen, inhalational induction was started with 2%–3% sevoflurane in 100% oxygen. The concentration of sevoflurane was increased gradually while preserving spontaneous breaths. After the adequate depth of anaesthesia was achieved, TruView-PCD (Picture Capture Device) VL (Truphatek International Ltd, Netanya, Israel) was inserted using left paraglossal approach. A Cormack Lehane (CL) IIb view was obtained with optimal external laryngeal manipulation. No other lesion near the laryngeal inlet was observed besides the cyst. We tried to insert a tube introducer but failed due to the large cyst with limited intraoral space and the procedure was abandoned. A nasopharyngeal airway was gently inserted in the left nostril and a breathing circuit was attached for pharyngeal insufflation of the oxygen/sevoflurane mixture through it. Nasotracheal intubation was

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Figure 1 Sequence of intubation in the infant with the vallecular cyst. (A) preoperative evaluation showing big intraoral cyst. (B) CT image showing the hypodense cyst at the base of the tongue. (C–E) Initial view with FOB. (F) Improved glottic view with FOB after the ‘three-person technique’. FOB, fibre-optic bronchoscopy.

then attempted through the right nostril using 4 mm internal diameter (ID) endotracheal tube (ETT) loaded on 2.8 mm FOB (model BF-XP290, Olympus Corporation). A big cyst reaching posterior pharynx was visualised and there was no space to advance the scope further. Jaw thrust was applied by an assistant while another person held the tongue in a gauze piece to apply vertical traction. These manoeuvres created a pharyngeal space posterior to the cyst for the advancement of the FOB beyond the cyst and allowed a partial view of the glottic aperture. Further manoeuvring of the scope allowed its advancement towards glottis and at this moment, 0.5 mL of 2% lignocaine was sprayed over the vocal cords through the working channel. The ETT was smoothly inserted into the trachea over FOB. Thereafter, 10 µg fentanyl and 2 mg atracurium were administered intravenously and GA was maintained with sevoflurane. The cyst wall and lining were then removed completely. Further intraoperative course was uneventful. The trachea was extubated at the end of the surgery and the baby shifted to high dependency unit for monitoring.

OUTCOME AND FOLLOW-UP

A histopathology examination confirmed the pathology to be a vallecular cyst. Following the procedure, the patient exhibited no signs of respiratory distress and feeding difficulties subsided.

DISCUSSION

A vallecular cyst is a rare but potentially life-threatening supraglottic airway pathology in infants. The presence of vallecular cysts in this vulnerable population can pose multiple challenges in airway management (box 1).

Narrower and collapsible airways of infants predispose them to early ventilatory compromise with any such airway lesion.⁷ Furthermore, they can exert a ‘ball-valve’ effect further aggravating the situation.⁸ Use of GA to facilitate intubation can, therefore, lead to complete airway obstruction (CAO), hypoxia and ventilatory failure.¹ A 2-month-old child with vallecular cyst developed CAO after failed attempts at direct laryngoscopy and required emergent surgical tracheostomy.³ Tracheostomy relieved CAO but the child developed negative pressure pulmonary oedema and could be decannulated only after 2 weeks. In another report, induction of GA in an 8-week-old infant with undiagnosed large vallecular cyst resulted in difficulty in

Box 1 The airway management challenges in an infant with a vallecular cyst

- ▶ Paediatric age group with the associated concerns of lack of cooperative and inadequate physiological reserves.
- ▶ Presence of airway obstruction and imminent respiratory failure.
- ▶ Sharing of the airway with the surgeon.
- ▶ Expected difficulty in mask ventilation and intubation.
- ▶ Possibility of cyst rupture and subsequent aspiration with any kind of transoral instrumentation.
- ▶ Associated conditions like laryngomalacia increasing the airway challenges.
- ▶ The poor general condition of the child due to feeding difficulties, dehydration and failure to thrive.

bag-mask ventilation and glottic visualisation. The airway could be secured after rapid aspiration of the cyst contents.⁹ However, even needle aspiration of cyst contents would require GA and there is a risk of aspiration of its contents. Also, it may be difficult to excise the mass after its aspiration. In our case, en masse removal of the cyst was planned. Cyst aspiration and tracheostomy were our backup plans in case of failure.

Considering the large size of the lesion, laryngeal mask airway may not be a feasible option if CAO develops after anaesthesia induction.⁴ Hence, the safest bet in such cases would be awake intubation. Awake intubation was not considered in our case because the child was uncooperative. Awake FOB can be quite challenging in this age group because of lack of co-operation, limitation on the dose of local anaesthetics for topicalisation and may have to be guided by transillumination and expiratory air bubbles.^{4–10} Also, in the presence of CAO, attempts at awake intubation can be very distressing. In a previous such report, awake tracheal intubation was attempted but required conversion to GA with a muscle relaxant.⁴

We chose inhalational anaesthesia with preserved spontaneous respiration and airway topicalisation to maximise safety and smoothen the process. Administration of a muscle relaxant in such cases can potentially lead to CAO by decreasing laryngeal tone. The other advantages of avoiding relaxants include decreased soft tissue collapse, improved view of the larynx and more procedure time. Nevertheless, inadequate depth can result in laryngospasm, coughing, displacement of the FOB and loss of the airway.¹¹

We had kept left paraglossal VL-guided intubation as plan A, as VLs provide non-line of sight view, have a slimmer profile than direct laryngoscopes and require less time than FOB.^{12–13} VL provide a better glottic view in difficult airway scenarios, help to quickly evaluate the extent of glottic obstruction and rule out any other pathology. We could indeed get a CL IIb view but due to inadequate space to manoeuvre the bougie and risk of cyst rupture, the procedure was abandoned and plan B (FOB under inhalational induction) was considered. We were unable to negotiate the FOB initially through the oropharynx due to the giant size of the cyst. ‘Three-person technique’ proved efficacious wherein, one person was operating the FOB, another person applied jaw thrust and the third one held the tongue with a gauze piece to apply vertical traction. These manoeuvres helped in navigating the FOB behind the lesion to visualise the glottic aperture. In an analogous previous report of an infant with a vallecular cyst, difficult nasal FOB was circumvented by a tongue stitch that helped to apply traction over the tongue

Table 1 Summary of the airway management techniques described in the literature in infants with a vallecular cyst

Technique	Author	Age (days)	Intub.	Difficult BMV	Anaesthetic agent/s	Vent.	Complications
DL	Cheng <i>et al</i> ⁴	105	Oral	No	Thiopentone, Sch	IPPV	Application of BURP, stylet-assisted intubation
	Schwartz <i>et al</i> ¹⁴	2	Oral	No	Sevoflurane, propofol	Spon	Case misdiagnosed as laryngomalacia
Paraglossal DL	Cheng <i>et al</i> ⁴	50	Oral	Yes	Halothane, Sch	IPPV	Initial attempt at awake DL failed
		65	Oral	No	Halothane, Sch	IPPV	Uneventful right paraglossal intubation
	Kalra <i>et al</i> ⁶	2	Oral	No	Sevoflurane	Spon	Initial failure conventional midline DL attempt
Cyst aspiration	Reiersen <i>et al</i> ⁷	56	Oral	Yes	NM	NM	Cyst aspirated with an 18G needle before intubation
	Cheng <i>et al</i> ⁴	53	Oral	Yes	TL, Sch Ketamine	CICO	DL failed and emergency cyst aspiration done
FOB	Cheng <i>et al</i> ⁴	58	Nasal	No	TL	Spon	Difficult FOB in an awake child, guided by indirect signs
		42	Nasal	No	TL	Spont	Direct visualisation not possible and FOB guided by indirect methods
	Wong <i>et al</i> ⁵	26	Nasal	No	Ketamine	Spont	DL, awake oral and nasal FOB failed, use of tongue traction suture
Tracheostomy	Batra <i>et al</i> ³	90	—	Yes	Halothane, propofol	IPPV	DL failed, NPPE and hypoxia developed

BMV, bag-mask ventilation; BURP, backward, upward, and rightward pressure; CICO, cannot intubate, cannot oxygenate; FOB, fibre-optic bronchoscope; IPPV, intermittent positive pressure ventilation; Intub.: intubation; NM, not mentioned; NPPE, negative pressure pulmonary oedema; Sch, succinylcholine; Spont, Spontaneous; TL, topical lignocaine; Vent, ventilation mode.

and improve glottic visualisation.⁵ The review of literature for airway management in patients with vallecular cyst suggests that aspiration of cyst is an effective rescue technique, maintenance of spontaneous respiration was preferred whereas awake intubation remains a difficult technique in this age group.^{4,7,14} Paraglossal technique of laryngoscopy can be effective in selective cases with minimal airway obstruction (table 1).

In this case, we had anticipated the airway management problems, formulated the primary, secondary and rescue plans and discussed the same beforehand with the parents and the surgeon.

Learning points

- Meticulous airway assessment, preparation, planning of primary and alternative airway management approaches using available logistics and preservation of spontaneous respiration are the keys to success in such cases.
- Para-glossal videolaryngoscopy can be a useful alternative to fibre-optic-assisted intubation in unilateral lesions for airway evaluation as well as securing the airway.
- The three-person technique described here can be useful to improve the view in such cases with supraglottic lesions obstructing the laryngeal aperture.

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