

Neonatal lingual gastric duplication cyst: A rare case report

K. V. Satish Kumar, Manoj Joshi, Naveen Vishwanath, Tanveer Akhtar, S. N. Oak

Department of Paediatric Surgery, BYL Nair Hospital and T. N. Medical College, Mumbai

Correspondence: Sanjay Oak, A / 11, 404, Neelam Nagar (Phase II), Mulund (E), Mumbai - 400 081, India.

ABSTRACT

Enteric duplication cysts occurring in tongue is rare. They present with feeding difficulty and rarely with respiratory distress. A one day old male baby presented with swelling in the oral cavity, interfering with feeding and inability to close the mouth. Under general anesthesia, subtotal excision was done and histopathology was gastric duplication cyst of fundic variety.

KEY WORDS: Lingual gastric duplication, enterocystoma, gastric heterotopia, foregut duplication cyst, lingual choristoma

Gastro-Intestinal duplications can occur anywhere from mouth to anal cavity, but one occurring in tongue in a neonate, is very rare. First published by Duncan and Daniel in 1942(2), this probably will be the 10th case in literature being reported. They occur due to entrapped endodermal cells during development. They usually present in the neonatal period and surgical excision is curative. We present a case of duplication cyst of tongue, which was excised sub totally and the baby is on follow up for recurrence.

CASE REPORT

A 1 day neonate, weighing 2.5 Kg, was brought with swelling in the oral cavity, with difficulty in feeding. Examination showed that the baby's mouth was wide open, due to a 4x5 cms cystic non-transilluminant mass occupying the entire oral cavity [Figure 1]. The origin of the swelling was difficult to ascertain, but was occupying the ventral aspect of tongue and pushing the dorsum of tongue upwards towards the palate. Clinically, a provisional diagnosis of mucus retention cyst or lymphangioma, was made. Pre-operative sonography of the swelling revealed a multiloculated cystic swelling with septations, probably suggestive of lymphangioma or a ranula. Surgical excision of cyst was planned. Under general anesthesia with awake orotracheal intubation, which was difficult as anticipated, sub mucosal partial excision of cyst was done, taking care to avoid injury to the sublingual salivary gland ducts. The cyst was arising from the ventral surface of the tongue, involving part of the lingual musculature and extending posteriorly, probably beyond base of tongue and hence it couldn't be

completely excised. Cut section showed it to be a multilocular cyst with turbid fluid. The baby was extubated on table and postoperatively it was maintained on naso-gastric feeds. Oral feeds were started on the 6th day. Histopathology of the lesion revealed it to be a multicystic structure, composed of columnar epithelial lining of gastric fundus variety, with smooth muscle and connective tissue, suggestive of gastric duplication cyst [Figure 2]. On follow-up after 2 months, the baby is doing well and there has been no recurrence.

DISCUSSION

Gastro-Intestinal duplication, is a true congenital anomaly. Alimentary tract duplications are cystic or tubular structures, lined by normal gastrointestinal



Figure 1: Photograph showing a neonate with open mouth due to a cyst in the tongue

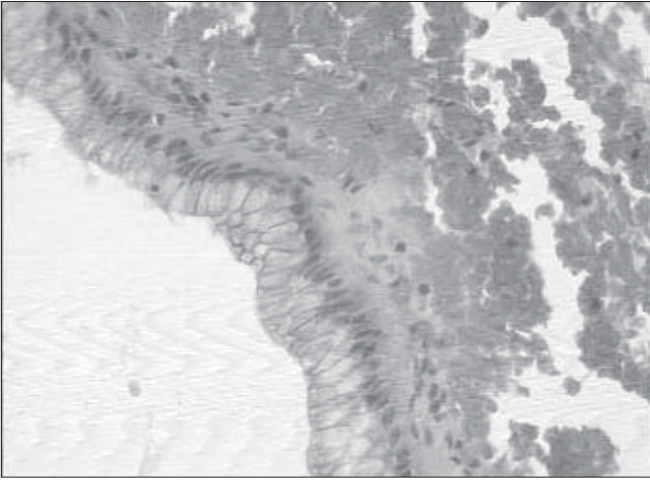


Figure 2: Histopathology under high power showing columnar gastric epithelium of fundic variety and smooth muscle

mucosa.^[1] These can occur anywhere from mouth to anus, the ileum being the commonest site. The criteria for duplication are - they are firmly attached to part of alimentary tract, have a well developed blood supply, often sharing with its normal viscus and their epithelial lining always resembles some part of the alimentary tract, most often, the adjacent viscus. First published by Duncan and Daniel in 1942, foregut duplications occurring in tongue are quite rare, with very few cases reported in literature.^[2] They can be lined by gastric, intestinal, colonic, or respiratory epithelium. Lingual foregut duplications arise from endodermal cells that become trapped during the process of fusion of lateral lingual swelling (distal tongue bud) and tuberculum impar (median tongue bud), in a 3-4 mm embryo. Various synonyms do exist, like enterocystomas, choristomas, gastric heterotopia, etc. In the tongue, enteric duplications in 60% of cases reported, occurring in the anterior 2/3rd.^[2] They can present from day one of life to any age and have also been discovered prenatally.^[3,4] They can be monolocular or multilocular cysts. They are usually seen in the neonatal period with enlarged tongue, interfering with feeding and rarely interfering with breathing.^[5] These lesions are more common in males.^[2] Rarely they can get infected, develop hemorrhage, or rupture causing asphyxia.^[2] The cyst can also ulcerate and may present as a sinus due to presence of parietal cells.^[6] Evaluation of the neonate should include high resolution ultrasound with doppler, in which a

nonvascular unilocular or multilocular cyst, would be seen. CT scan and MRI scan are useful in older children and adults, but may be difficult in neonates, as sedation may lead to respiratory compromise.^[2] Differential diagnoses include lymphangioma, ranula, hamartoma, teratoma, dermoid cyst, etc.

Ideally one should go for complete surgical excision as it is the definitive treatment and this should not be delayed, as respiratory compromise is imminent.^[5] Oro-tracheal Intubation in these babies is difficult, as retraction of tongue and visualization of glottis is hampered, because of lack of space in the oral cavity and hence, the need for Naso-tracheal intubation or tracheostomy may arise.^[7] Excision of the cyst should be complete and can be accomplished using cautery or laser. Cases are reported, where having diagnosed prenatally, ex-utero intrapartum (EXIT) treatment has been done.^[7,8] If the child presents acutely with respiratory difficulty, emergency aspiration of the cyst^[7] with subsequent excision at a later date, can be done. If completely excised, the cosmetic and functional outcome is good.^[9]

REFERENCES

1. Willner A, Feghali J, Bassila M. An enteric duplication cyst occurring in the anterior two-thirds of the tongue. *Int J Pediatr Otorhinolaryngol* 1991;21:169-77.
2. el-Bitar MA, Milmoe G, Kumar S. Intralingual foregut duplication cyst in a newborn. *Ear Nose Throat J* 2003;82:454-6
3. Rousseau T, Coureur S, Senet-Lacombe E, Durand C, Justrabo E, Malka G, *et al.* Prenatal diagnosis of enteric duplication cyst of tongue. *Prenat Diagn* 2004;24:98-100.
4. Hall NJ, Ade-Ajayi N, Peebles D, Pierro A. Antenatally diagnosed duplication cyst of the tongue: Modern imaging modalities assist perinatal management. *Pediatr Surg Int* 2005;21:289-91.
5. Ameh EA, Jimoh AO, Rafindadi AH, Shehu SM. Sublingual gastric duplication cyst causing respiratory obstruction: Case report. *East Afr Med J* 2000;77:394-5.
6. Parikh DH, Ibrahim SK, Cook RC. Peptic ulceration in a lingual sinus. *J Pediatr Surg* 1991;26:99-100.
7. Chen MK, Gross E, Lobe TE. Perinatal management of enteric duplication cysts of the tongue. *Am J Perinatol* 1997;14:161-3.
8. Kong K Walker P, Cassey J, O'Callaghan S. Foregut duplication cyst arising in the floor of mouth. *Int J Pediatr Otorhinolaryngol* 2004;68:827-30.
9. Wiersma R, Hadley GP, Bosenberg AT, Chrystal VJ. Intralingual cysts of foregut origin. *J Pediatr Surg* 1992;27:1404-6.
10. Eaton D, Billings K, Timmons C, Booth T, Biavati MJ. Congenital Foregut Duplication Cysts of the Anterior Tongue. *Arch Otolaryngol Head Neck Surg* 2001;127:1484-7.