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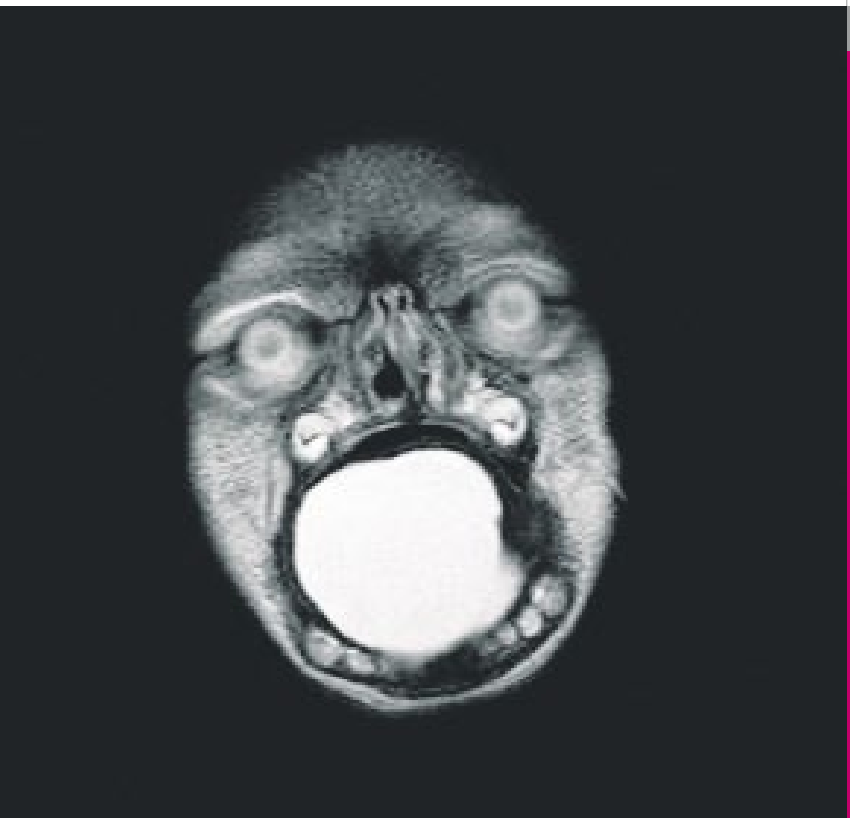
Peripartal management of a prenatally diagnosed large oral cyst



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In this patient, fetal ultrasound examination performed in the 23rd week of gestation revealed a cystic lesion within the oral cavity. The mother's medical and obstetrical history were unremarkable. There was no family history of any congenital anomalies. Follow-up ultrasound examinations showed progressive increase in cyst size until the fetus was no longer able to close his mouth (Fig. 1). Since there was no polyhydramnios the fetus apparently was still able to swallow amniotic fluid.

The mother was hospitalized at 39 1/7 weeks of gestation, one day before a planned elective caesarean section. Because of non-reassuring fetal heart tracing, an emergency caesarean section was performed on the day of admission. Delivery was attended by neonatologists, a pediatric surgeon and a pediatric anesthetist to guarantee different options of airway management.

A baby boy was delivered with an Apgar score of 7, 9, and 9 at 1, 5, and 10 minutes, respectively. His birth weight was 2290 g ($P < 3$). Respiratory distress became apparent within a few minutes of birth. A large cyst filled and obstructed the oral cavity (Fig. 2). It was possible to bag-mask ventilate the baby, and following sedation the larynx could be visualized. After muscle relaxation, he was intubated orally at 15 minutes of life without difficulty (Fig. 3). A few minutes later, the endotracheal tube was changed

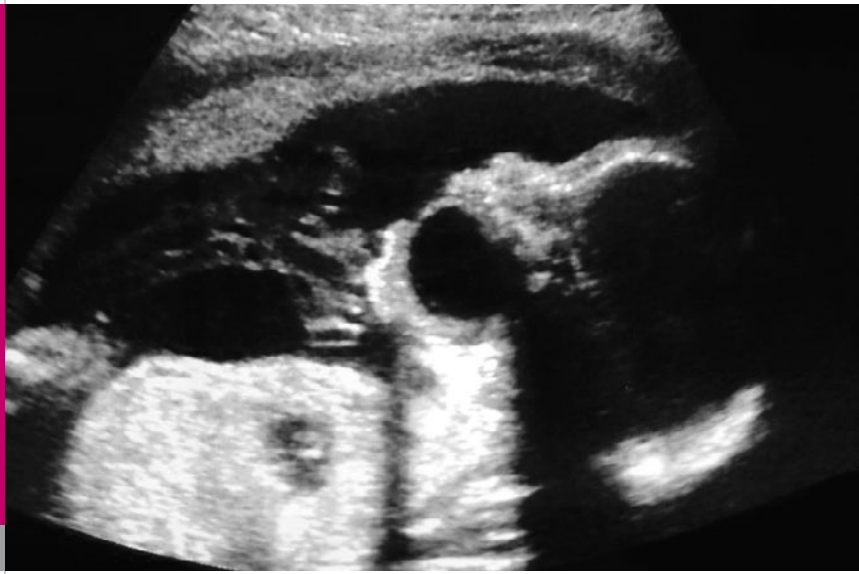
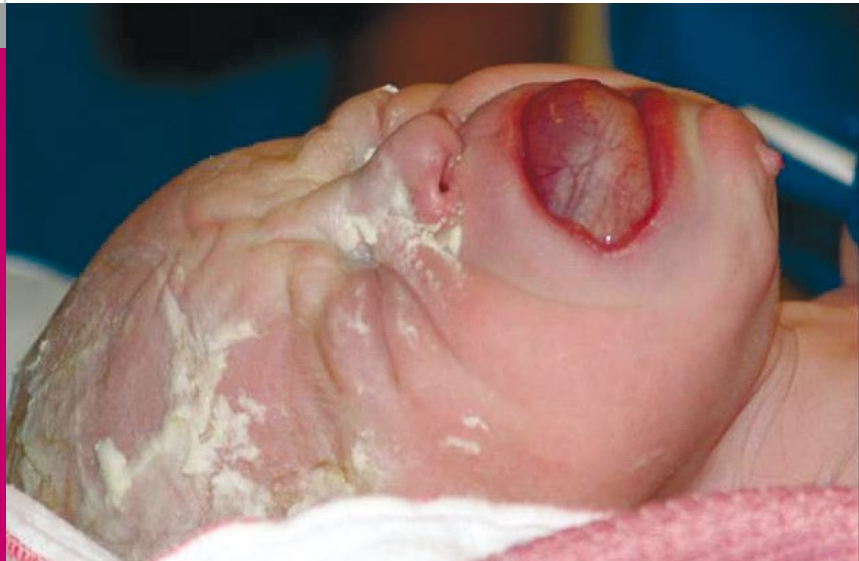


Fig. 1

Prenatal ultrsound outlining a large oral cyst.

Appearance of the cyst protruding from the mouth immediately after birth.

Fig. 2



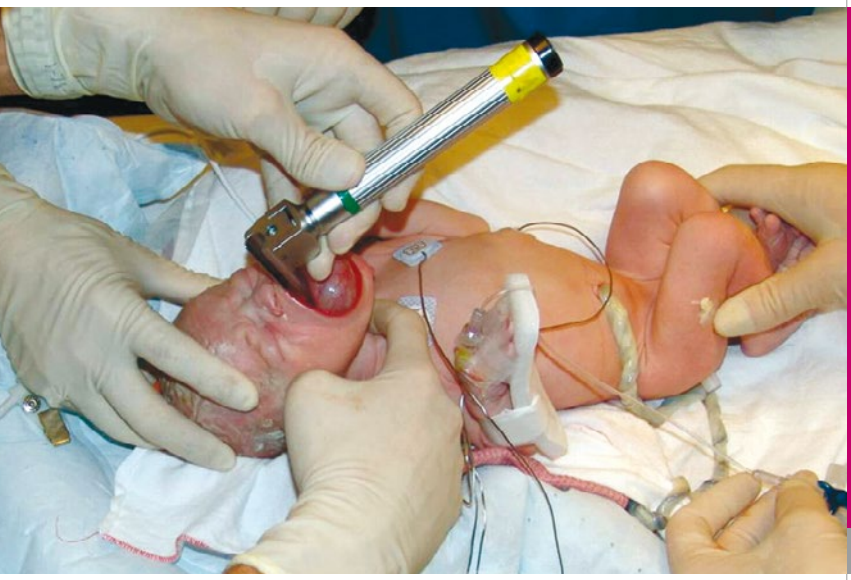


Fig. 3

Visualization of the vocal cords following sedation.

Appearance of the cyst after nasotracheal intubation; note small skin tag at the tip of the chin.

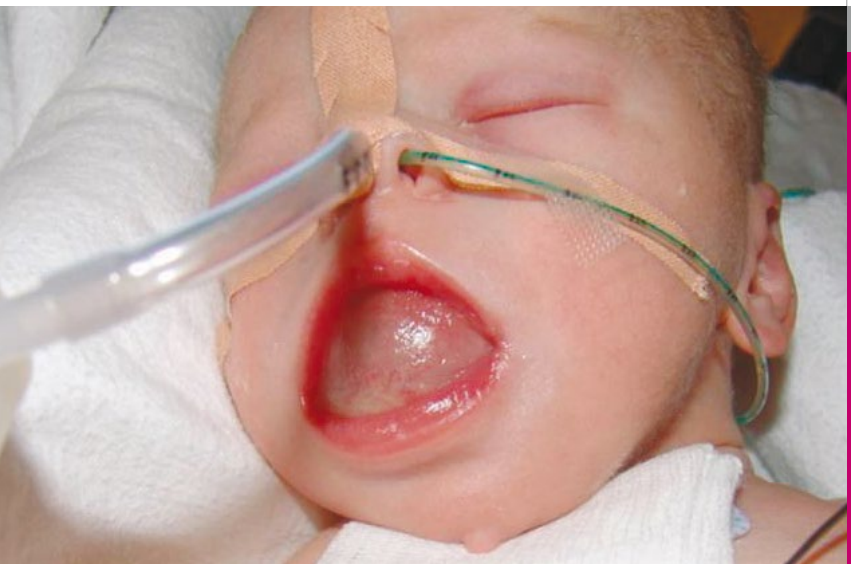


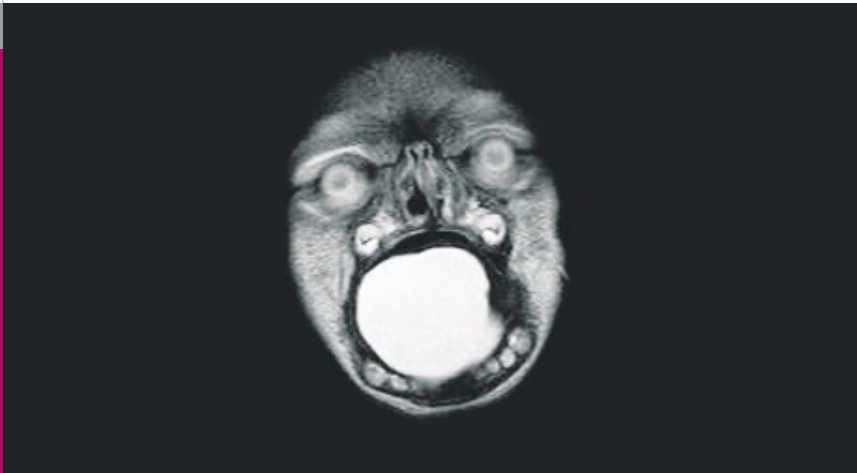
Fig. 4

to the nasotracheal position (Fig. 4). Lung compliance appeared normal and he could be ventilated easily without supplemental oxygen.

Apart from a small skin tag at the tip of his chin (Fig. 4), no other malformations were identified. On the second day of life, an MRI study more clearly outlined size (3.8 cm x 3.6 cm x 3.5 cm) and localization of the cyst (Fig. 5, 6). Secondary to the pressure his maxillary teeth were shifted ventrally. Complete surgical resection of the cyst was performed electively on day three of life (Fig. 7). He was extubated immediately following the procedure and was breathing comfortably without any respiratory support. On histology, there was a cystic lesion outlined predominantly by ciliated and mucous producing respiratory epithelium as well as single groups of mucinous glands surrounded by edematous stroma and striated muscle (Fig. 8, 9). No residual thyroid tissue was recognizable.

T2-weighted coronal MR image showing the large fluid filled cyst.

Fig. 5



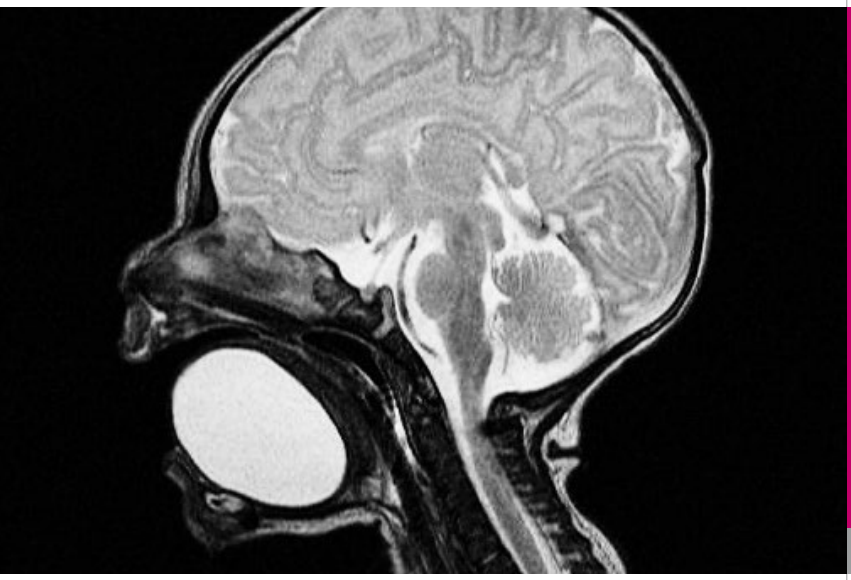


Fig. 6

T2-weighted sagital MR image outlining size an position of the cyst.

Intraoperative appearance of the cyst.

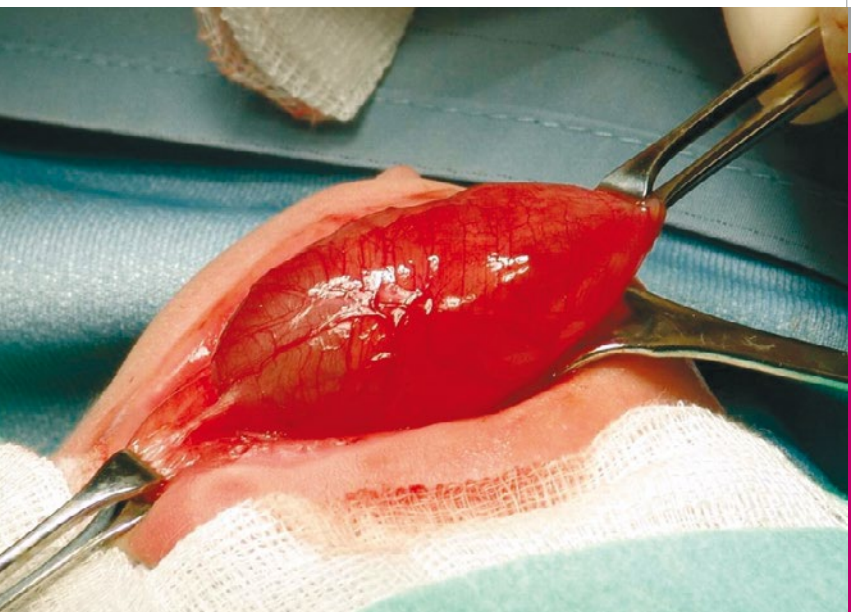


Fig. 7

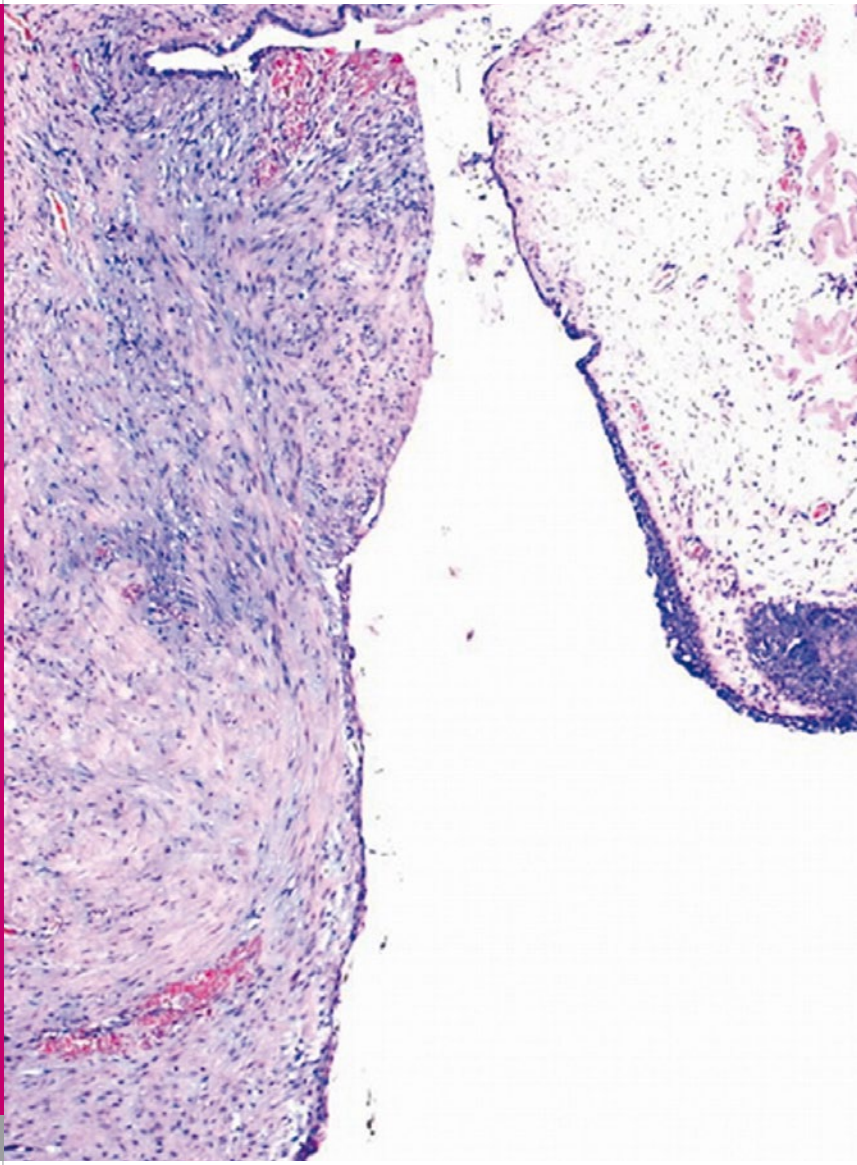
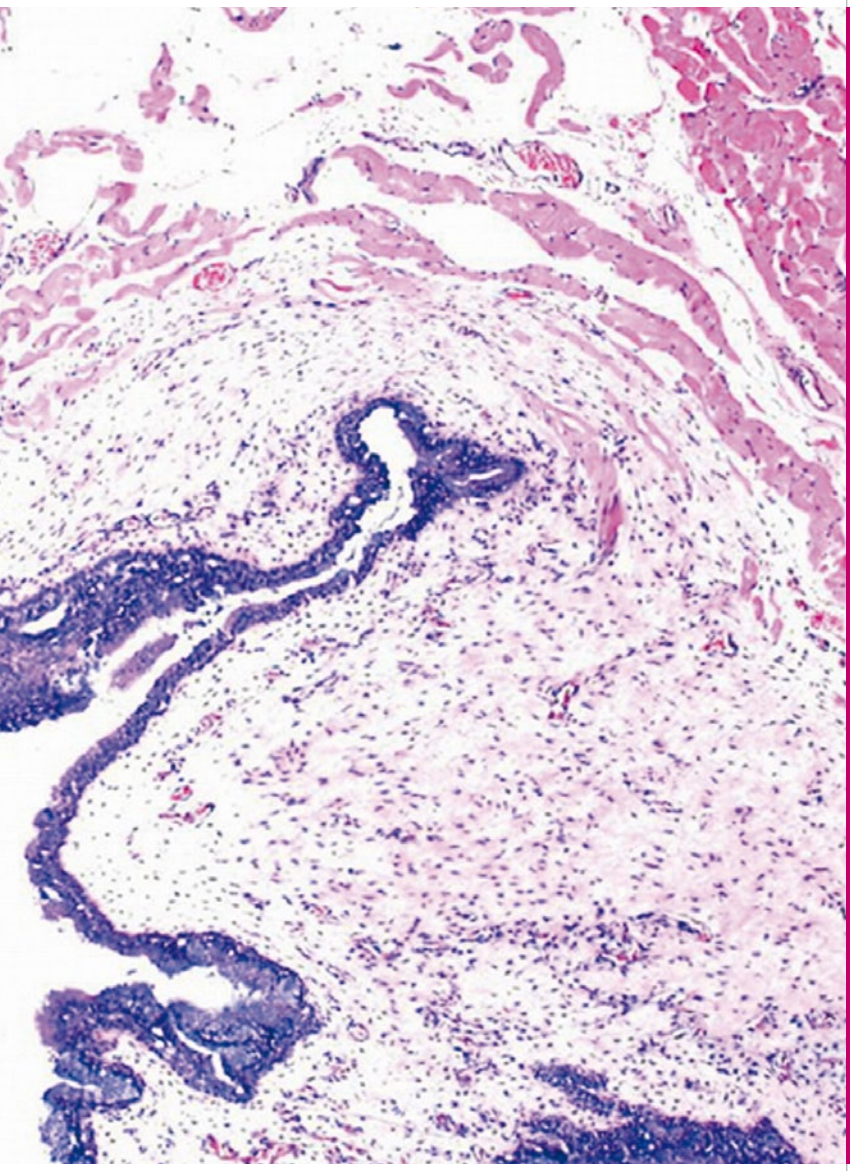


Fig. 8

Histology: cystic lesion surrounded by edematous stroma and striated muscle.



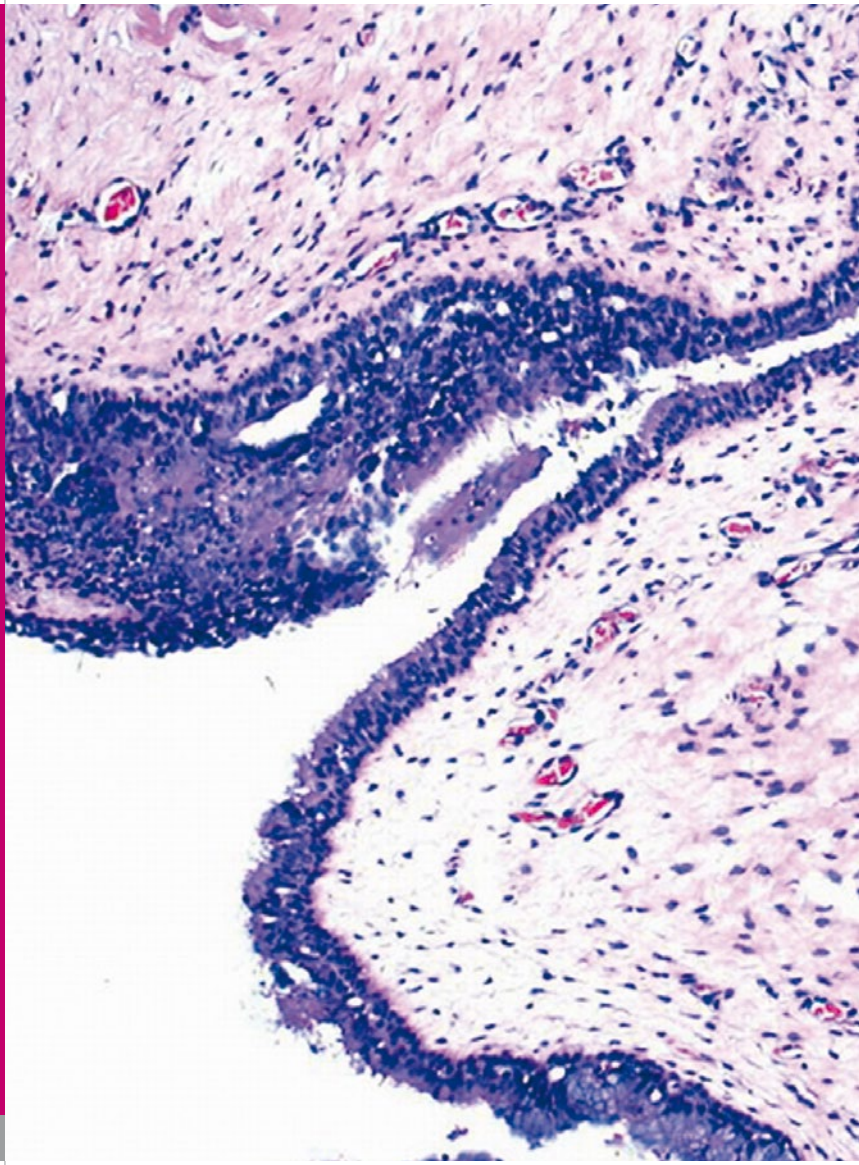
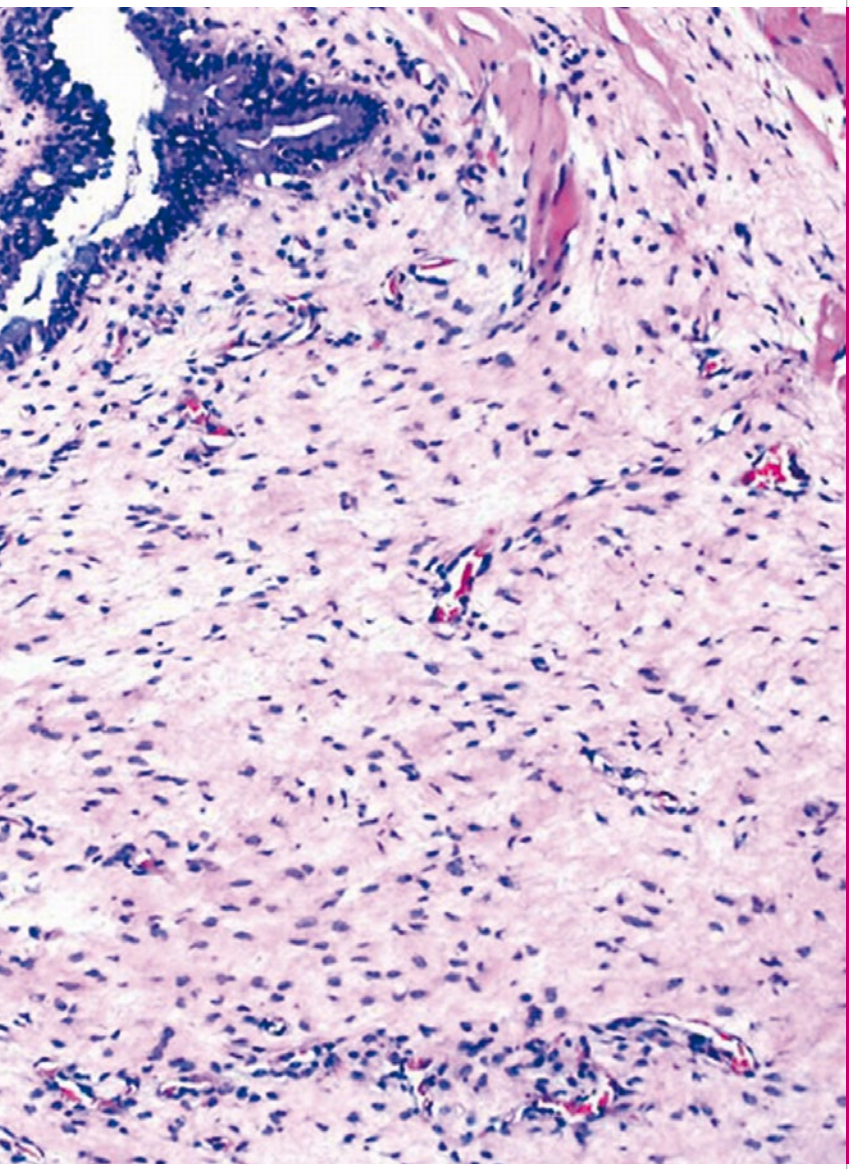


Fig. 9

Histology: ciliated, pseudostratified columnar epithelium.



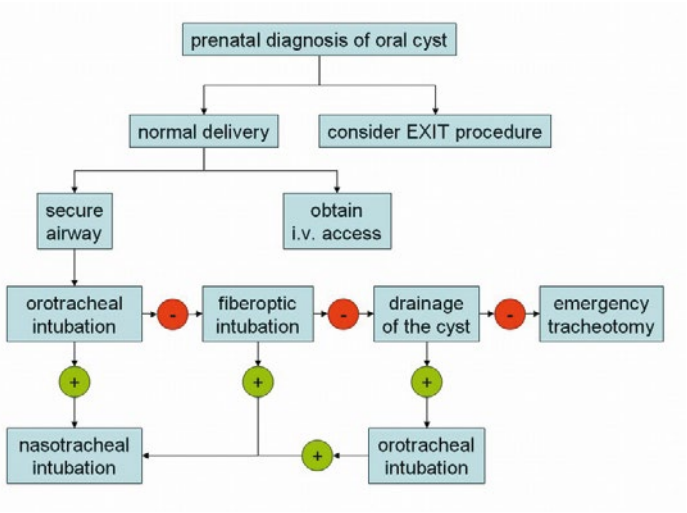


Fig. 10

Suggested management algorithm for prenatally diagnosed large oral cysts.

Congenital oral and tongue lesions are rare in the perinatal period. Although most of them are benign, their localization may cause severe upper airway obstruction immediately after birth. The differential diagnosis of these lesions has previously been described in the Case of the Month 10/2005. When such an abnormality is detected during routine antenatal screening, interdisciplinary perinatal discussions should focus on the treatment options to ensure upper airway patency immediately following birth. Risks (especially severe uterine hemorrhage) and benefits of an EXIT procedure (Ex Utero Intrapartum Treatment) have been described in the literature and should be weighed against alternative airway management strategies after delivery. Because of lack of experience at our institution, we opted against an EXIT procedure. Delivery was attended by a multidisciplinary team to provide support for fiberoptic intubation, cyst drainage or emergency tracheotomy should conventional airway management fail. We propose an algorithm for the management of prenatally diagnosed obstructive oral cysts (Fig. 10).

REFERENCES

1. Karam O et al. Congenital sublingual cyst. www.neonet.ch: Case of the Month 10/2005

2. Busuttil M et al. Congenital laryngeal cyst: benefits of prenatal diagnosis and multidisciplinary perinatal management. Fetal Diagn Ther 2004;19:373-376

3. Hall NJ et al. Antenatally diagnosed duplication cyst of the tongue: modern imaging modalities assist perinatal management. Pediatr Surg Int 2005;2:289-291

4. Dahlgren G et al. Four cases of the ex utero intrapartum treatment (EXIT) procedure: anesthetic implications. Int J Obstet Anesth 2004;13:178-182

5. Lalwani AK, Engel TL. Teratoma of the tongue: a case report and review of the literature. Int J Pediatr Otorhinolaryngol 1992;24:261-268

6. De Backer A et al. Strategy for management of newborns with cervical teratoma. J Perinat Med 2004;32:500-508

7. Shih GH et al. The EXIT procedure facilitates delivery of an infant with a pretracheal teratoma. Anesthesiology 1998;89:1573-1575

8. Hirose S et al. The ex utero intrapartum treatment procedure: Looking back at the EXIT. J Pediatr Surg 2004;39:375-380

9. Chen MK et al. Perinatal management of enteric duplication cysts of the tongue. Am J Perinatol 1997;14:161-163

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