CASE REPORT

Congenital mandibular epulis – A rare oral lesion in a newborn

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Abstract

Introduction: Congenital epulis or congenital granular cell tumour is a rare benign lesion arising from alveolar mucosa of neonates which can cause respiratory and feeding problems. It is also known as Neumann's tumour. Females are affected 10 times more frequent.

Method: A review of medical record was performed.

Result: A newborn presented with lesion over left lower gum which was seen during antenatal abdominal ultrasonography scan by Obstetrics team at 35-week of gestation. The case was referred for ex utero intrapartum treatment procedure in view of anticipating upper airway obstruction. Upon delivery via Caesarean section, patient had no obstructive symptoms however was immediately intubated few hours of life to secure airway. Computed tomography scan confirmed a soft tissue mass arising from either lower alveolus or tip of tongue. The lesion was excised and histopathological examination reported as congenital epulis.

Conclusion: Even though it is a rare case, congenital epulis should be considered as one of differential diagnosis of an oral mass. A multi-disciplinary team management is important in view of possibility of upper airway obstruction post delivery.

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1. Introduction

Congenital epulis is a benign congenital granular cell tumour which was first described by Neumann in 1871.1 The tumour has a smooth surface, arises from alveolar mucosa and varying in size from several millimetres to a few centimetres which is typically protruding from the neonate’s mouth. Congenital
epulis occur more frequently in the maxilla as compared to the mandible with the ratio of 3:1. The lesion is mainly on its anterior portion which can lead to feeding difficulty and airway obstruction. Surgical excision is a treatment of choice immediately after detection because spontaneous regression is rare and high possibility of airway obstruction as well as feeding difficulties.

2. Case report

A 20-year-old lady G1P0 was referred by Obstetrics team with a suspected mass seen at fetus’s mouth during a routine antenatal ultrasonography. At 35-week gestation, the trans-abdominal ultrasonography showed a mass measuring $28 \times 24 \times 20$ mm seen in the fetus’ mouth. The mass was well-demarcated and homogeneous in nature. It was predicted to arise from just posterior to the fetus’s lower lip. There was no increased vascularity in the mass and the tongue can be seen separately posterior to the mass. Because of concern for disruption of the mass or mechanical obstruction of the airway during a vaginal delivery, a lower segment Caesarean section (LSCS) with ex utero intrapartum treatment (EXIT) was planned.

At 38-week gestation, LSCS delivery was performed with a multi-disciplinary team of specialists. The delivery was conducted under general anaesthesia. When the fetal head was delivered, the newborn cried immediately, confirming at patent oral airway and the EXIT procedure was abandoned. The newborn underwent endotracheal intubation without difficulty and was moved to an adjacent operating room, where the mass was inspected. The lower lip was intact and not communicating with the mass. It was a lower alveolar mass with rubbery consistency measuring 2 cm in diameter (Fig. 1). No other abnormalities were noted. The newborn was planned for emergency computed tomography (CT) scan to assess the extension of the mass and planning for excision biopsy. The newborn was self-extubated on 5 h of life however was not re-intubated in view of no sign and symptoms of upper airway obstruction.

CT scan done with findings of rounded homogenously enhancing soft tissue mass measuring $1.8 \times 2.1 \times 1.8$ cm (Fig. 2). The lesion had no clear plane of demarcation with lower lips and tip of tongue with suspicious erosion to the adjacent lower alveolus. There was no calcification or fat component within. The pharynx and nasal cavity were patent. It was concluded that the soft tissue mass is likely arising from the lower alveolus.

Excision was done at day-13 of life with finding of $2 \times 2 \times 1$ cm and firm broad-based mass arising from lower alveolus (Fig. 3a). Excision was done using monopolar diathermy and hemostasis secured (Fig. 3b). The neonate was admitted to the neonatal intensive care unit and safely extubated after the surgery and feeding began on the next day. Histological examination confirmed the diagnosis of congenital epulis. Microscopic description shows a polypoidal tissue which was covered by skin. The underlying tissue is composed of granular cells which are large, polygonal to spindle in shape. Nuclei are small and bland. Many capillary blood vessel seen, however there was no evidence of malignancy. The recovery of both the mother and neonate were uneventful and they were transferred back to the initial referring hospital.

3. Discussion

Congenital granular cell tumor commonly referred as congenital epulis, is a rare lesion in the oral cavity which exclusively encountered in newborns. The typical appearance is smooth, pedunculated and pink mass. Congenital epulis differs from other oral soft tissue tumours by its maxillary or mandibular alveolar location (3:1) usually around the canine incisor, its female predominance (8:1) and it’s solitary in nature. Tumour in the oral cavity potentially can cause feeding difficulty and upper airway obstruction.

It is important for the clinicians to be aware of more serious differential diagnoses of an oral lesion in the newborns. These include the rhabdomyosarcoma, and osteogenic and chondrogenic sarcomas which can all have catastrophic consequences. Treatment modalities for these lesions are very different, and early diagnosis and treatment are essential. However, provisional diagnosis is often made clinically at birth due to typical occurrence in the alveolar mucosa of newborn girls and is confirmed histologically. This is applied in this case. Prenatal ultrasound is complementary and can narrow the differential diagnosis of oral masses.

The histogenesis of congenital epulis is not certain however it is thought to be non-neoplastic, degenerative or reactive lesion as this lesion self-regress with absence of local recurrence even after incomplete excision. The origin could be from odontogenic epithelium, undifferentiated mesenchymal cells, pericytes, fibroblast, smooth muscle cells, nerve-related cells and histiocytes. Recent immunohistochemical staining and ultrastructural examination favour myofibroblasts as the cells of origin. There is a report suggested that it is a neuroectodermal derivation. Presence of autophagosomes which contain collagen precursors may theorize the tumor cells represent early mesodermal cells that express pericytic and myofibroblastic feature that undergo cytoplasmic autophagocytosis.

When the lesion is large or interfering with feeding and breathing, the treatment will be simple surgical excision. Radical excision is discouraged as it can damage underlying structures. There was no case of recurrence reported even with incomplete excision. There was no malignant transformation reported as well. Epulis has been reported to regress spontaneously after birth, suggesting that its growth is dependent on maternal hormonal milieu associated with

![Figure 1](image_url) Oral mass seen upon delivering via caesarean section.
pregnancy. Although spontaneous regression can take place, it cannot be expected. A large or multiple congenital epulis may cause feeding problems, respiratory obstruction or interfere with normal development of jaws and teeth. In this case, the lesion was interfering with breast-feeding and hence early removal was seemingly necessary.

It is important to diagnose neonatal congenital epulis antenatally mainly by using ultrasonography. However there was a case which does not reveal any abnormality at first trimester which suggests that the tumour may have become apparent at later stage. The earliest reported case was detected at 26-week gestation. In our case, the tumour became apparent in the ultrasonography at 35-week gestation. Therefore, a weekly serial antenatal ultrasonography in the third trimester can be suggested to rule out any possibility of tumour in neonates.

By doing prenatal imaging, the choice of delivery method can be decided. Large tumour may compromise a normal vaginal delivery and a Caesarean section may be necessary. The earlier identification of a tumour in a fetus also can assist to the intervention planning. This will allow the family to prepare psychologically for procedures that will take place and its possible risk. However, if the lesion is only detected upon delivery, it is important to ensure the airway secured and maintained before any invention is going to take place.

The management of congenital epulis is of multidisciplinary interest as many professionals are involved with the prenatal and delivery care. Good interaction and planning between professionals involved with prenatal care and those involved in the treatment of the lesion will optimize the diagnosis and treatment approaches.

4. Conclusion

Congenital epulis is a rare oral mass that present at birth in newborns which most likely develops late in utero. Although the lesion is visually impressive and distressing, it is a benign lesion. Prompt surgical treatment is necessary to avoid complications such as airway obstruction or feeding difficulty. Prenatal imaging plays an important role to detect such lesions for further planning on mode of delivery and further management on neonates.

Conflict of interest

None declared.

References


