

Congenital epulis: A rare benign tumour

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SUMMARY

Congenital epulis is a rare benign pedunculated tumour of the oral cavity arising from the alveolar ridges. It is usually detected in newborns and can be successfully resected surgically. We report a case of a newborn baby who had a 5x3x3cm pedunculated lobar mass arising from the upper alveolar ridge.

KEY WORDS:

Congenital epulis; newborn; benign tumour

INTRODUCTION

Congenital epulis is a rare benign tumour arising from the alveolar ridges of newborns. They are mainly composed of mesenchymal tissue and are usually surgically resected. They can pose quite a surprise to clinicians' when presented at childbirth, as it is usually quite large and difficult to detect through prenatal ultrasound.¹ In large tumours, it can cause problems with swallowing, breastfeeding and respiratory compromise. Treatment is generally straightforward, the recovery period is unremarkable and the risk of recurrence is low.²

CASE REPORT

An otherwise healthy newborn girl was referred immediately after birth for a large pedunculated mass protruding from her oral cavity. The baby was born at term via caesarean section weighing 2.7kg and was breathing spontaneously with no distress. She had normal antenatal scans.

Physical examination revealed a 5x3x3cm pedunculated mass arising just right of midline of the upper alveolar ridge. The mass was not obstructing the airway but did interfere with breastfeeding (Figure 1a). There were no dysmorphic features of note.

As there were no other medical or congenital problems detected, a decision was made to excise this mass before allowing the patient to be discharged home. This was performed under local anesthesia in the operating theatre. After injecting 0.5% Marcain (with 1:100000 adrenaline) into the stalk, the lesion was ligated with a 2/0 silk suture and the mass was excised via monopolar diathermy (Figure 1b). No feeding vessel was noted in the stalk. Hemorrhage was minimal. Postoperative recovery was uneventful. The patient

was breastfeeding immediately after and discharged the next day.

Follow-up at two weeks revealed a pristine looking alveolar bed with no evidence of the stalk. The patient was noted to be feeding well, thriving and gaining weight at the six-month mark.

Histological examination revealed an encapsulated lesion covered with squamous epithelium macroscopically (Figure 2a). Microscopically there was evidence of granular eosinophilic and basophilic cytoplasm centrally located nuclei consistent with findings of a congenital epulis (Figure 2b).

DISCUSSION

Congenital epulis was first described by Neumann in 1871 and is a rare benign tumour of the jaw that occurs exclusively in neonates. Epulis literally means "of the gums" in greek and the tumour usually arises from the gingiva of the future site of the maxillary canine or the lateral incisors, but the unerupted teeth are not involved.² The lesion occurs in females more than males with a ratio of 10:1 and is more common in the maxilla than the mandible (3:1).³ It usually presents as a pedicled smooth lobulated tumour with a firm consistency over the future site of the canine-incisor area.¹ Congenital epulis is reported to be an isolated finding without associated congenital abnormalities.⁴ It appears sporadically with no familial tendency and has an incidence rate of about 6 per million.²

Diagnosis/differentials

Differential diagnosis depends on site, size and velocity of growth. It is broad and based on clinical perspective only. These diagnoses could include congenital malformations, such as cephalocele and dermoid, as well as benign and malignant neoplasms, such as lymphatic malformation and rhabdomyosarcoma or non-specific soft tissue masses such as a fibroma or a vascular malformation or other developmental anomalies such as granular cell tumours (GCTs) or neuroectodermal tumours of infancy.^{1,4}

Imaging

The tumour can be diagnosed by prenatal ultrasound or with the more superior magnetic resonance imaging (MRI) as early as at 26 weeks of gestation or in third trimester of

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Fig. 1: A. Tumour on the day of birth B. Wound site at Day 1 post excision (printed with consent).

pregnancy.¹ However, it is important to remember that prenatal findings are non-specific and could include a myriad of differential diagnosis as stated above.⁴ Nonetheless, these modalities could be helpful in pre-operative planning for delivery. This is because large tumours may impact on vaginal delivery and a caesarean section might be indicated. Imaging with MRI could be helpful in confirming the diagnosis postnatally and ensuring that the lesion has no local extension prior to surgical resection.

Histology

The proposed source of origin of an epulis includes undifferentiated epithelial and mesenchymal cells, pericytic, and fibroblastic, histiocytes, nerve-related, smooth muscle, and myofibroblastic cells.¹ Therefore, there could be multiple differential diagnoses histologically with the closest one being GCTs, which has a risk of malignancy.⁴ It is therefore important to differentiate these tumours by their studying their immunohistochemical (IHC) profile and histomorphology. In congenital epulis, vimentin, which is a marker for intermediate filament in cells of mesenchymal origin is 100% positive and S-100 protein, which is a marker for neural cell origin is 100% negative.^{1,5} Histomorphologically, congenital epulis exhibit large round cells with granular, eosinophilic cytoplasm and small eccentric nuclei with a delicate fibrovascular network separating the cells with no cellular or nuclear pleomorphism as noted in our case.⁵ In GCTs however, there are pseudoepithelialomatous hyperplasia, more conspicuous nerve bundles and less vascularity compared to an epulis.¹

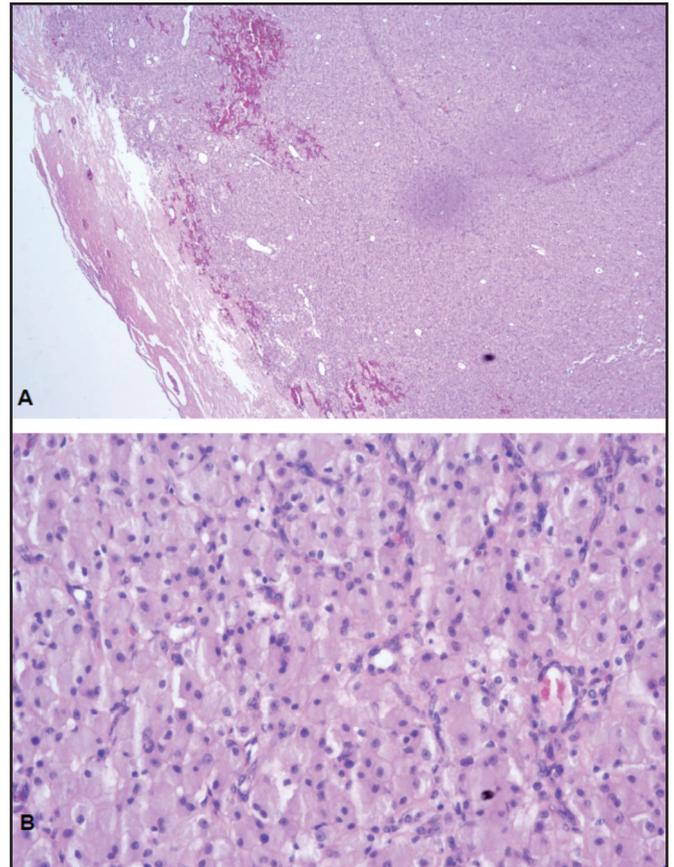


Fig. 2: A. Mass is covered by thin stratified squamous epithelial mucosa at low power view. Tumour is in sheets and tight clusters with intervening capillaries. B. High power view shows plump tumour cells with abundant granular eosinophilic cytoplasm. Nuclei is small and pyknotic with no atypia or increase mitosis.

Treatment

Spontaneous regression has been noted in the literature however simple surgical excision under local or general anesthesia is the mainstay of treatment.⁴ In our case above, we performed an early surgical excision under local anesthesia as it interfered with the patient's appearance and feeding. The second reason for early surgical intervention was to reduce the emotional burden of the parents before bringing the baby home. Of note, there has been no mention of recurrence or malignant transformation in the literature yet.^{1,2}

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