

Congenital epulis: a report of two cases and review of the literature

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ABSTRACT

Background: Congenital epulis of the newborn is a rare benign soft-tissue tumour of the gingival, which is also called gingival granular cell tumour of the newborn. These slow growing soft tissue tumors affect the gingivae of a new born child. It is essential to document the presentation and the management of this lesion because of its rare nature. **Aim:** To highlight the clinical presentation, diagnosis and management of the lesion. **Methods:** Two clinical cases of congenital epulis were used to illustrate the presentation and surgical management of the lesion. **Findings:** The first clinical case is a one month old female child with a protruding gum tumour on the anterior alveolar process of the mandible. The second case is a one week old male child with a gingival growth on the anterior alveolar process of the mandible. All the tumours were surgically excised under local anaesthesia. **Conclusion:** Congenital epulis could interfere with the functions of the oral cavity. Therefore, it is imperative to surgically excise it when diagnosed. Following the management of the two cases presented, it is essential that the mouth of all newborns must be examined to rule out any such lesion.

Key words: Congenital epulis, gingival, tumour, alveolar process, neonate, surgical excision

INTRODUCTION

Congenital epulis is a rare benign soft-tissue tumour of the gingival origin occurring in the neonate.^[1] It is also referred to as a gingival granular cell tumour of the newborn.^[1] This tumour was first described by Neumann in 1871,

more than 170 cases have been reported since then.^[2] This soft tissue tumour is smooth-surfaced, pedunculated, and sometimes lobulated and the diameter ranges from few millimeters to 9mm.^[3] It is usually located on the gingival and found predominantly on the maxillae.^[3] It is predominant in females with a ratio of male to female of 1:4.^[3] Fuhr and



Krogh^[4] reported that the tumour occurred eight times more in females than males and three times more often on the maxillae than the mandible.^[3] Although benign by its size, it induces breathing and feeding problems.^[3]

Congenital epulis usually presents as a pedunculated, non-ulcerated, reddish pink mass of varying sizes, however, multiple lesions may also occur in the same or different alveolar ridges.^[4]

"Epulis is a Greek term literally meaning 'of the gums' and is used to describe a wide variety of gum lesions, regardless of their pathological origin".^[2,5] Histologically, congenital epulis shows significant similarity with granular cell tumours (GCTs) which is commoner.^[2,5] Epilus are however solely in the neonate with typical location, plexiform arrangement of capillaries, and lack of pseudoepitheliomatous hyperplasia.^[4] GCTs are ubiquitous neoplasms that occur in all age groups, and rarely affect the gingival with occasionally malignant transformation.^[4]

"Histological, congenital epulis shows characteristically large cells with granular cytoplasm and spindle cells resembling fibroblasts".^[4] The lesion is benign and no recurrence or metastasis has been reported. The aim of this report is to document the presentation and management method of this gingival tumour found in neonates. This is a report of two cases of congenital epulis in a newborn and a one-month old baby.

CASE REPORT

CASE ONE

A one month old female baby referred to our Dental Clinic from a paediatrician from a General Hospital in the same city. History obtained from the mother showed that at birth, there was a small swelling of the gum of the anterior alveolar process of the mandible, which was painless and was growing progressively. The mother presented in the hospital when the tumour prevents the baby from feeding properly. The baby is a product of term, uneventful pregnancy. Examination of the tumour showed a single, large, oblong, lobulated and

pedunculated swelling on the anterior alveolar ridge of the mandible on the left side (figure 1). The surface of the swelling was smooth, reddish in colour and showed a prominent blood vessel at the base of the tumour. The large size of the tumour was causing feeding problems for the baby. A clinical diagnosis of congenital epulis was made. Full blood count of the baby shows haemoglobin concentration of 11g/dl and white blood cell count of 4×10^6 . The patient vital signs shows: temperature of 36.7°C , pulse rate of 90cycle/minute and respiratory rate of 22/min. The tumour was excised after infiltration was done with 2% lignocaine with 1:10,000 adrenaline, local anaesthesia and a blood vessel underneath the tumour was ligated to stop the bleeding after excision. The tumour measured 3cm x 4cm x 5cm (figure 2).



Figure 1: One month old baby with a protruding gum tumour on the alveolar process of the mandible

It was completely excised from the base and examined histopathologically. Histology showed overlying stratified squamous epithelium and vascular stroma, sheets and clusters of cells with abundance of granular eosinophilic cytoplasm and small uniform nuclei, along with some myxoid, haemorrhagic and ulcerated areas. This confirmed the diagnosis of congenital epulis (figure 3). The patient was seen for review after 7 days when the suture was removed. The healing was satisfactory and the feeding habit has normalized.



Figure 2: The excised pedunculated tumour

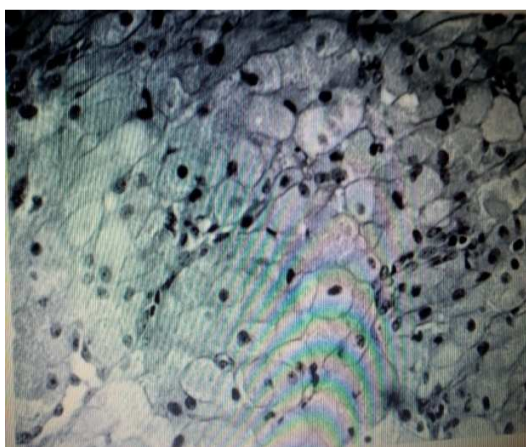


Figure 3: Histograph of congenital epulis showing numerous granular cells in cytoplasmic stroma

CASE TWO

This case is a one week old baby boy referred from the paediatric department of the hospital to the Dental Clinic. The child was born with a tumour on the left anterior alveolar process of the mandible. The baby is a product of full term normal pregnancy. The delivery of the baby came through normal vaginal delivery. There was no positive history of drug usage during pregnancy. Clinical examination shows a 1cm x1cm x1.6cm pedunculated, smooth surfaced and protruding mass into the oral cavity, regular pink coloured soft tissue growth on the alveolar crest of the left side of the mandible (figure 4). The tumour poses feeding challenges for the baby. Full blood count of the baby shows

haemoglobin concentration of 10g/dl and white blood cell count of 4×10^6 .

The tumour was excised after infiltration with 2% lidocaine with 1in 10,000 adrenaline local anaesthetic agent. The haemorrhage was minimal, direct pressure on the alveolar process was used to achieve haemostasis. Histology revealed overlying stratified squamous epithelium and vascular stroma, a large polyhedric cells with vacuolar central nuclei and eosinophilic granular cytoplasm and a diagnosis of congenital epulis (figure 4). A week review showed satisfactory healing of surgical site.



Figure 4: Appearance of a small gum mass arising from alveolar process of the mandible

DISCUSSION

Congenital epulis is a rare benign neonatal tumour. About 167 cases of this benign lesion was reported before 1993.^[5] It is usually seen in neonates, although prenatal diagnosis with ultrasound has been documented at 26 weeks gestation.^[6] The tumour is commonly seen over the incisor-canine region of the maxilla.^[7] The two cases reported occurred in the mandible, but also at the incisor-canine region. The maxillary and mandibular alveolar ridges have been reported to be simultaneously involved in about 10% cases.^[8] The cases reported occurred only on the mandible. The tumour usually interfere with feeding.^[5] The two cases reported interfere with the feeding of the babies. The clinical presentation consists of a lobular, or ovoid or sessile or pedunculated swelling

covered by a smooth mucosal surface usually in the maxillae. "The diagnosis is usually made on clinical grounds alone, although difficulties may arise when the size of the lesion is small, or the index of suspicion is low".^[6] Dental lamina cyst of alveolar process could be misdiagnosed as congenital epulis. These lesions behave in a benign manner and no recurrent or metastatic lesions have been reported.

To the best of our knowledge, the incidence of this lesion has not been documented. A facility in the USA reported only two cases over the period of 21 years.^[5] In University Hospital of Wales, a tertiary referral centre for Otolaryngology and Neonatology, no case of this benign lesion has been reported since 1980.^[5]

The appearance of the lesion by postnatal ultrasound and MRI has been documented.^[8] MRI shows the gingival origin of the lesion without local extension.^[9] Management is with surgical excision, although spontaneous regression has been documented.^[10] The two cases reported were surgically excised.

The actual origin of the lesion is still unclear. The tumour is said to be mesenchymal in origin.^[2] GCTs are reported to arise from schwann cells, thus show strong reactivity to S-100 protein.^[2] Different theories of the origin of congenital epulis has been postulated, these include myoblastic, neurogenic, odontogenic, fibroblastic, and histocytic theories.^[3] Lack *et al.*^[10] reported congenital epulis to be reactive in origin. It has been reported to occur primarily in neonates, and more in females.^[9] This suggests a hormonal mechanism in the aetiology. Some studies have shown no evidence of either oestrogen or progesterone receptors, and as such suggest an alternative histogenesis.^[6,7] Vered *et al.*^[9] in their review of 33 lesions concluded that the immunohistochemical profile does not implicate any specific cell types for the histogenetic origin of the benign lesion. There has not been any reported case of recurrence after excision.^[11] The differential diagnosis include: haemangioma, lymphangioma, fibroma, granuloma, rhabdomyosarcoma and heterotropic gastrointestinal cysts.^[12,13] Three months post operative review of the cases reported showed no recurrence. Congenital epulis of the newborn must be excised when

diagnosed to allow the baby access to good meals.

CONCLUSION

It is essential that adequate attention be paid to oral examination of the newborn so as to diagnose this lesion before it starts to affect oral functions.

RECOMMENDATION

Midwives, gynaecologists and paediatricians need to always do routine oral examinations for all newborns for early diagnosis of this gum tumour.

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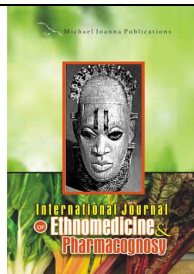
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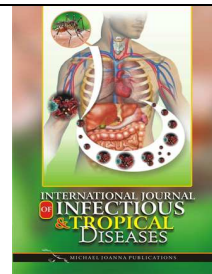
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