

# Congenital Epulis of the Newborn: A Report of Three Cases and Review of the Literature

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ABSTRACT

**Objective:** 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. This slow growing soft tissue tumour affect commonly the labial side of 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. The objective of this study was to highlight the clinical presentation, diagnosis and management of the lesion.

**Methods:** Three clinical cases of congenital epulis were used to illustrate the presentation and surgical management of the lesion.

**Results:** The first clinical case was a one month old female child with a protruding gum tumour on the anterior alveolar process of the maxillae. The second case was a one week old male child with a gingival growth on the anterior alveolar process of the maxillae. The third case was new born male child with the tumour at the anterior alveolar process of the maxillae. All the tumours were surgically excised under local anesthesia.

**Conclusion:** Congenital epulis of the newborn could interfere with the functions of the oral cavity. Therefore, it is imperative to surgically excise it when diagnosed. Sequel to the management of the three cases presented, it is necessary to always examine the oral cavity of the new born to rule out the presence of epulis and any other oral lesion.

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## INTRODUCTION

Congenital epulis is a rare benign soft-tissue tumour of the gingival origin occurring in the neonate. It is also referred to as a gingival granular cell tumour of the newborn (Neumann, 1871). This tumour was first described by Neumann in 1871, more than 170 cases have been reported since then (Lapid et al., 2001). This soft tissue tumour is smooth surfaced, pedunculated or sessile, non-ulcerated, reddish pink mass of varying sizes, sometimes lobulated and the diameter ranges from few millimeters to gmm. However, multiple lesions may also occur in the same or different alveolar ridges (Kanotra et al., 2006). It is usually located on the gingivae and found predominantly on the maxilla and in females with a ratio of male to female of 1:4

(Kanotra et al 2006). The tumour has been reported to occur eight times more in females than males and three times more on the maxilla than the mandible (Fuhr & Krogh, 1973). Although benign in nature, larger lesions can cause breathing and feeding problems (Kanotra et al., 2006).

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Histological, congenital epulis shows characteristically large cells with granular cytoplasm and spindle cells resembling fibroblasts (Fuhr & Krogh, 1973). The lesion is

benign and no recurrence or metastasis has been reported (Sliva et al., 2007).

Congenital epulis shows significant histological similarity with granular cell tumors (GCTs) which is commoner (Kanotra et al., 2006, Fuhr & Krogh, 1973). However, it is differentiated from GCTs because it is solely seen in the neonate with typical location, plexiform arrangement of capillaries, and lack of pseudoepitheliomatous hyperplasia (Kanotra et al., 2006, Fuhr & Krogh, 1973). GCTs are ubiquitous neoplasms that occur in all age groups, and rarely affect the gingival with occasionally malignant transformation (Fuhr & Krogh, 1973). The objective of this report is to document the presentation and management of maxillary congenital epulis.

## CASE SERIES

### CASE ONE

A one month old female baby was referred to our Dental Clinic from a Pediatrician from a General Hospital in the same city. History obtained from the mother showed that at birth, there was a small swelling of the gingiva in the anterior alveolar process of the maxillae, which was painless and was growing progressively. The mother brought the baby to hospital when the tumour prevented the baby from feeding properly. History obtained from the mother revealed that the baby was a product of term and uneventful pregnancy. Examination of the

tumour showed a single, large, oblong, lobulated and pedunculated swelling on the anterior alveolar ridge of the maxillae on the left side. The surface of the swelling was smooth, reddish in colour and showed a prominent blood vessel at the base of the tumour (Figure 1A). A clinical diagnosis of congenital epulis was made. Full blood count of the baby shows haemoglobin concentration of 11g/dl, white blood cell count of  $4 \times 10^5$  and body weight of 6.5kg. The patient vital signs showed: temperature of 36.7°C, pulse rate of 90 cycle/minute and respiratory rate of 22/min. The tumour was completely excised from the base 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 2cm x 4cm x 6cm (Figure 1B).

The excised tissue was sent for histology. Histology showed overlying stratified squamous epithelium and vascular stromal, sheets and clusters of cells with abundance of granular eosinophilia cytoplasm and small uniform nuclei, along with some myxoid, haemorrhagic and ulcerated areas. This confirmed the diagnosis of congenital epulis (Figure 1C). The patient was seen for review after 7 days when the suture was removed. The healing was satisfactory and feeding habit has normalized.

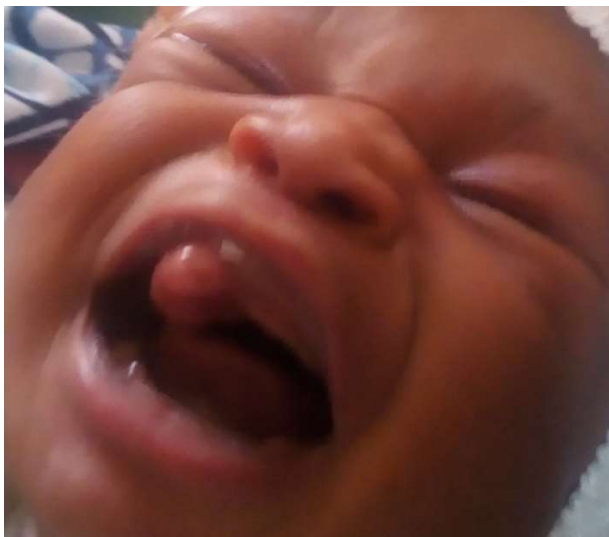


Figure 1A: A 1month old female baby with the tumour on the anterior alveolar process of maxillae.



Figure 1B. The excised tumour.

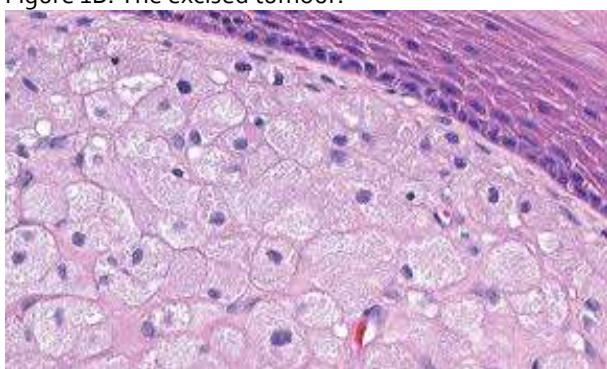


Figure 1C: Histograph of congenital epulis showing numerous granular cells in cytoplasmic stroma.



Figure 2: A week old baby boy with the tumour on the anterior alveolar process of the maxilla

## 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 maxillae. The baby was 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 2cm x 1cm x 4cm sized, 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 maxillae (Fig. 2). The tumour poses feeding challenges for the baby. Full blood count of the baby shows haemoglobin concentration of 10g/dl, white blood cell count of  $4 \times 10^6$  and body weight of 3.5kg. The tumour was excised after infiltration with 2% lidocaine with 1 in 10,000 adrenaline local anaesthetic agents. 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 polyhedral cells with vacuolar central nuclei and eosinophil granular cytoplasm and a diagnosis of congenital epulis. A week review showed satisfactory healing of surgical site.

### CASE THREE

A day old male child was referred to our Dental Clinic from the maternity ward of our hospital.

History obtained from the mother revealed that the baby was a product of term and uneventful pregnancy. Examination of the oral cavity showed a single, large, oblong, multi-lobulated and pedunculated gum mass in the anterior alveolar process of the maxillae, which was firm and not tender to palpation. The surface of the swelling was smooth, reddish in colour and the tumour measured 2cm x 2cm x 3cm (Figure 3). A clinical diagnosis of congenital epulis was made



Figure 3: A day old male child with the tumour at the anterior alveolar process the maxilla

TABLE 1: TABULAR FORM OF THE REPORTED CASES

CASES	1	2	3
Age	1month	1week	1day
Sex	Female	Male	Male
Location	Anterior maxilla	Anterior maxilla	Anterior maxilla
Referral	General Hospital	Pediatrics BDTH	Maternity BDTH
Antenatal History	Good care	Good care	Good care
Gestation History	Full term	Full term	Full term
Drug use in pregnancy	None	None	None
Mode of Delivery	Vaginal delivery	Vaginal delivery	Vaginal delivery
Gross feature	Oblong, sessile	Pedunculated	Lobulated
Size	2cmx4cmx6cm	1cmx1.6cmx2cm	2cmx2cmx3cm
HB	11g/dl	10g/dl	10g/dl
Clinical Diagnosis	Congenital epulis	Congenital epulis	Congenital epulis
Histological Findings	Granular cytoplasm	Large polyhedral cells	Eosinophilia cytoplasm
Procedure	Excision	Excision	Excision
Treatment outcome	Satisfactory	Satisfactory	Satisfactory

## DISCUSSION

Congenital epulis is a rare benign neonatal tumor (Neumann, 1871). About 167 cases of this benign lesion were reported before 1993. It is usually seen in neonates, although prenatal diagnosis with ultrasound has been documented at 26 weeks gestation (Lapid et al., 2001). The tumour is commonly seen over the incisor-canine region of the maxilla (Kanotra et al., 2006). All the three cases reported occurred in the anterior alveolar process of the maxillae. The maxillary and mandibular alveolar ridges have been reported to be simultaneously involved in about 10% cases (Fuhr & Krogn, 1973). The tumour usually interferes with feeding (Lapid et al., 2001). The three cases reported interfered with the feeding of the babies. The clinical presentation consists of a lobular or ovoid, sessile or pedunculated swelling covered by a smooth mucosal surface usually in the maxillae (Sliva et al., 2007). Cases 1 and 2 were sessile, while 3 was multi-lobulated and pedunculated lesion. The diagnosis is usually made on clinical grounds alone, although difficulties may arise when the size of the lesion is small as the index of suspicion is low (Lack et al., 1982). Dental lamina cyst of alveolar process could be misdiagnosed as congenital epulis, except that previous contain cystic fluid. The two lesions behave in a benign manner and no recurrent or metastatic lesion has been reported (Lack et al., 1982).

The actual origin of the lesion is still unclear, but was said to be mesenchymal origin (Fuhr & Krogn, 1973). Granular Cell Tumours are reported to arise from Schwann cells, thus show strong reactivity to S100 protein. Different theories of the origin of congenital epulis had been postulated, these include myoblasts, neurogenic, odontogenic, fibroblastic, and histolytic theories. Congenital epulis was reported to be reactive in origin (Lack et al., 2001). It has been reported to occur primarily in neonates, and more in females (Lack et al., 1982). All the cases in this study occurred in neonates but two were males and one was a female. This may suggest a hormonal mechanism in the aetiology. Some studies have shown no evidence of either oestrogen or progesterone receptors, and as such suggested an alternative histogenesis (Sliva et al., 2007). However, in a review of 33 lesions it was concluded that the immuno-histochemical profile does not implicate

any specific cell types for the histogenetic origin of the benign lesion (Vered et al., 2009). 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 (Nakata et al., 2002). In University Hospital of Wales, a tertiary referral Centre for Otolaryngology and Neonatology, no case of this benign lesion has been reported since 1980 (Zerener et al., 2013).

The appearance of the lesion by postnatal ultrasound and magnetic resonance imaging has been documented (Raissaki et al., 2005). MRI shows the gingival origin of the lesion without local extension (Raissaki et al., 2005). Management is surgical excision of the tumour, although spontaneous regression has been documented (Lack et al., 1982). The three cases reported were surgically excised.

There has not been any reported case of recurrence after excision (Zucker & Buenecha, 1993, Kusakawa et al., 1997). The differential diagnosis includes: Dental lamina cyst, haemangioma, lymphangioma, fibroma, granuloma, rhabdomyosarcoma and heterotopic gastrointestinal cysts (Bosanquet & Roblin, 2009). 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 nutritive 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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### Conflict of interest

The authors declare that they have no conflicts of interest.

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