

# Congenital Epulis: The first reported case in Cameroon

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## Abstract

Congenital epulis (CE) is a rare benign tumor that arises from the gum pads of neonates especially on the anterior maxillary regions and in females. It can be obstructive and impair with feeding and respiration. We report the case of congenital epulis in a female neonate arising from both the maxillary and mandibular areas. Diagnosis was clinically suspected before surgical resection and confirmed on histology. Recovery and follow up was uneventful and no recurrence or malignant degeneration has been noted.

## INTRODUCTION

Congenital epulis also called congenital granular cell tumour of the newborn is a rare benign gingival cell tumor that most commonly occurs along the anterior alveolar ridges of a neonate.<sup>1,2,3,4,5</sup> The tumor is very rare and was described for the first time by Neumann in 1871 and so is also called Neumann's tumor.<sup>1,6,7</sup>

Congenital epulis presents as a mass protruding through the mouth of a neonate; interfering with respiration and/or feeding depending on the size. The presence of this lesion in a newborn can be traumatizing to the parents.

The social and esthetic problems arising from this pathology, the simplicity of diagnosis and treatment with an excellent cure rate is the reason for this case report from the Yaounde Gynaeco-Obstetric and Pediatric Hospital and the first reported case in Cameroon.

## CASE REPORT

A 28 year old primiparous woman underwent an elective caesarian section on the 28/07/2008 for materno-fetal dystocia at 41 weeks of gestation.

Past history of pregnancy was uneventful. Prenatal ultrasound was done in the first, second and third trimester and were all normal.

The infant was female and the Apgar score was 8- 10- 10 at the 1<sup>st</sup>, 5<sup>th</sup> and 10<sup>th</sup> minute respectively and weighed 3400gms.

Physical examination revealed two separate pedunculated gingival masses protruding and almost completely obstructing the mouth (Figure 1). The masses were irregular, multilobular and erythematous. The larger mass multilobular and arose from the right latero-anterior maxillary (5cmx3cm) alveolar ridge and the smaller was located on the right anterior mandibular alveolar ridge (2.5cmx2cm). Both were attached onto the gums through two small fibrous peduncles. The size of the masses prevented normal closure of the infant's mouth hindering normal breast or bottle feeding. However the masses did not obstruct the nostrils so there was no respiratory distress. The bony maxillary and mandibular structures and the tongue were all normal. No other abnormal findings were noted.

**Figure 1**

Figure 1: Presentation of the tumor at birth



Laboratory work-up, and chest and abdominal imaging did not reveal any abnormalities.

The baby was fed via a 6F nasogastric feeding tube. The masses did not increase in size and remained vascularised. Surgical excision for histology of the masses was decided and done at the 10<sup>th</sup> day. This was done after ligation (with an absorbable suture) at the base of the two pedicles under local anaesthesia with 2% lidocaine. Bleeding was minimal and the post operative period was uneventful. The site of incision healed within few a days. The feeding tube was removed and baby started feeding normally as from the next day.

**Figure 2**

Figure 2: Mouth of baby at the immediate post operative period.



The excised masses were sent for histological examination. The histology showed a well differentiated malpighian epithelium on a richly vascularised connective tissue containing histiocytes, lymphocytes and plasmocytes. There were no malignant cells.

The baby was seen one week later and there were no abnormalities (Figure 3). She was then seen at 1, 4 and 6 months on an outpatient basis, and there were no signs of relapse.

**Figure 3**

Figure 3: Mouth of baby one week later.



**DISCUSSION**

Congenital epulis (CE) is a rare benign gingival tumor that occurs on the gum pads of a neonate. It is a congenital granular cell lesion (CGCL) having a close resemblance to granular cell myeloblastoma. The word “epulis” previously described for the lesion is derived from the Greek word and means “on the gums” or “gum boil”.<sup>5</sup> The term “CE” is preferred to “CGCT” because the latter suggests a neoplastic origin, not applicable to these lesions. Moreover, CE does not represent a variant of granular cell tumors.<sup>7</sup> It differs from other granular cell tumors in adults by its exclusive origin from the neonatal gingiva, the scattered presence of odontogenic epithelium, the more elaborate vasculature and lack of interstitial cells with angulate bodies.<sup>12</sup>

C.E. only occurs on the gum pads of neonates while granular cell tumors can occur any where in the body and at any age.

A protruding mass from the mouth of a newborn can be very distressing for the parents and interfere with feeding and/or breathing of the infant. The case we report was the first child of a young couple. The parents were so distressed because the paternal grandparents almost put an end to their marriage since they believed that the baby was bewitched.

Since its discovery by Neumann in 1871, it has also been termed Neumann’s tumor, and very few cases have been reported worldwide.<sup>156</sup> To date fewer than 200 cases have been described worldwide.<sup>7</sup> No case been reported in Cameroon although a case of granular cell tumor in a 23 years old female has been reported.<sup>8</sup>

It is known to have a marked female preponderance with a girl – boy ratio of 8:1.<sup>123679</sup> The sex of our case goes further to support this finding. The reason for female dominance is not clear. However a hormonal stimulus has been suspected but no proved since no estrogen and progesterone receptors have been found on the tumor to support this hypothesis.<sup>2467</sup> The exact etiology is unknown. Several theories have been suggested, namely, myeloblastic, odontogenic, neurogenic, fibroblastic, histiocytic and endocrinologic.<sup>245</sup>

It arises from the mucosa of the gingiva and most commonly from the anterior part of the maxillary alveolar ridge. Maxillary predilection occurs in the ratio of 3:1.<sup>456</sup> They often occur as single lesions and in 10% of cases they may be multiple.<sup>6</sup> In our patient it was multiple and multilobular in both the maxillary and mandibular gum pads.

CE is not usually associated with abnormalities of the teeth or additional congenital malformations. They arise at the future site of the maxillary canine or the lateral incisors, but the unerupted teeth are not involved.<sup>2</sup> However, some authors have reported hypoplastic or absence of underlying tooth, mild maxilla hypoplasia and mildly deformed lips, flat nose with the absence of anterior nasal spine and reduced antero posterior dimension of maxilla.<sup>46</sup> These abnormalities are thought to be caused by the volume and weight of overlying mass preventing the normal growth of the affected tissue. In our patient, there was no deformation of the surrounding structures and there were no other associated malformations. Tooth eruption had occurred normally at 6 months.

Diagnosis of this lesion can be made before birth during routine obstetrical ultrasound as from the 25 weeks of pregnancy when the images of the fetal face reveal a homogenous well-circumscribed, non-septate oral mass with Doppler finding inconsistent with hemangioma.<sup>471011</sup> However some cases develop in late pregnancy and so are missed on early ultrasound.<sup>2</sup> Antenatal polyhydramnios can be caused by partial obstruction of the oral cavity and the resultant ineffective swallowing.<sup>411</sup> This was not observed in our patient whose antenatal ultrasounds was absolutely normal. It is also surprising that the abnormality was not seen during the third trimester obstetrical ultrasound despite its size.

Diagnosis in the neonate is usually not difficult because of its typical appearance and occurrence in the maxillary alveolar ridge in female neonates. The lesion is usually a pedunculated, non ulcerated pink mass of a few millimeters

to several centimeters. It takes the form of a pedicular tumor, with a smooth or lobular surface, and of firm, rubbery consistency. It has a pinkish-violet colour and inserts itself into the alveolar ridge of the dental arch through a pedicle of variable size.<sup>11</sup> The differential diagnosis of a large mass in the fetal or neonatal oral cavity should include encephalocele, dermoid cysts or teratomas, and benign and malignant neoplasms including hemangiomas, lymphatic malformations, melanotic or pigmented neuroectodermal tumors and rhabdomyosarcomas.<sup>12</sup>

Even with the advent of modern histopathological techniques, it has not been able to depict specific cellular features unique to this lesion.<sup>5</sup> Histologically, the lesions are composed of large cells with eosinophilic granular cytoplasm looking like histiocytes and fibroblasts resting on vascular fibrous connective tissue.

Histologically CE shows highly vascularised fibrous tissue with nests of polygonal cells with large clear and granular cytoplasm and a small nucleus, with a normal overlying epithelium.<sup>46</sup> Granular cells show antigenic features of neural crest and mesenchymal cells.<sup>12</sup> These features make it difficult to distinguish it from granular cell tumor by light microscopy alone. Immunohistochemical features of CE have less variability than GCT.

Immunohistochemically, CE stains positively for vimentin and sometimes to desmin, respectively markers for collagen and muscle cells, and negatively for specific macrophage markers (MAC 387, CD68), alpha fetoproteins, S-100 proteins and neuron-specific enolase (NSE). The later markers are diffusely present in granular cell tumors.<sup>71213</sup> This procedure is not feasible in our setting.

The recommended treatment is prompt surgical resection at the stalk under local or general anesthesia since they can obstruct breathing and feeding.<sup>2369</sup> Treatment abstention and follow up has been proposed by some authors for small lesions which do not pose feeding problems because spontaneous regression has been reported.<sup>314</sup> However the delay of treatment in our case was due to delay in diagnosis. The mass was feared to be either malignant or highly vascularised, but finally two surgical knots were put on the stalk of each mass before resection and then after sent for histology. Radical surgery is not necessary since neither recurrence, malignancy nor damage to future dentition has been reported.

## CONCLUSION

This case is a CE which presents as a benign mass protruding through the mouth of a new born baby and can be visually impressive and frightening to parents, and distressing to the baby. When large and obstructive it can impair feeding and respiration and prompt surgical treatment is warranted. The diagnosis is suspected clinically and treatment by simple surgical resection is curative and no recurrence has ever been reported.

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## References

1. Lapid O, Shaco-Levy R, Kierger Y, Kachko L, Sagi A. Congenital epulis. *Pediatrics* 2001Feb ; 107(2) ; e22.
2. McGuire TP, Freilich MM, Sandor GKB. Congenital epulis: A surprise in the neonate. *J Can Dent Assoc* 2006; 72(8):747-50.
3. Nouri S, Mahdhaoui N, Bouafsoun C, Zakhama R, Omezzine, Yaacoubi MT, et al. Salem N, Seboui H. Epulis congenital obstructive a double localization. A propos d'une observation chez un garçon. *Arch Ped* 2008; 15: 1083-6.
4. Koch BL, Meyer III C, Egelhoff JC; Congenital epulis. *AJNR* 1997 Apr; 18: 739-41.
5. Dash JK, Sahoo PK, Das SN. Congenital granular cell lesion "congenital epulis". Report of a case. *J Indian Soc Pedo Prev Dent* 2004 Jun; 22(2): 63-7.
6. Kannan SK, Rajesh R. Congenital epulis - congenital granular cell lesion: A case report. *J Indian Soc Pedod Prev Dent* 2006 Jun; 104-6.
7. Leocata P, Bifaretti G, Saltarelli S, Corbecelli A, Ventura L. Congenital (granular cell) epulis of the newborn: A case report with immunohistochemical study on the histogenesis. *Ann Saudi Med* 1999; 19(6):527-9.
8. Bengondo MC, Onana J, Sando Z, Ebolo X, Bengono G. Epulis géante pédiculé : A propos d'un cas. *Med Buccale Chir Buccale* 2006 ; 12(3) :159-62.
9. Subramaniam R, Shah R, Kapur V. Congenital epulis. *J Postgrad Med* 1993; 39(1): 36.
10. Metogho EM, Oury JF, François M, Collet M. Epulis congénitale. A propos d'un cas de diagnostic anténatal. *J Gynecol Obstet Biol Reprod* 2003 Dec ; 32(8-C1):6.
11. Thoma V, Idrissi B, Kohler M, Becmeur F, Viville B, Favre R. Prenatal diagnosis of congenital epulis : A case study. *Fetal Diagn Ther* 2006; 21: 321-5.
12. Takahashi H, Fujita S, Satoh H, Okabe H. Immunohistochemical study of congenital gingival granular cell tumor (congenital epulis). *J Oral Pathol Med* 1990, 19: 492-6.
13. Michel B, Couly G. Tumeurs et dysplasies tumorales de la cavité buccale du nouveau-né et du nourrisson. *EMC - Dentisterie* 2004 ;1 : 214-27.
14. Jenkins HR, Hill CM. Spontaneous regression of epulis of the newborn. *Arch Dis Child* 1989; 64(1):145-7.

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