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Congenital epulis in a newborn – a case report in Benin City, Nigeria

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ABSTRACT

Introduction and aim. Congenital epulis is a rare benign tumor that affects the oral cavity of newborns which typically presents as a solitary mass on the maxillary alveolar bridge at birth, with a predilection for the female gender. The rarity of congenital epulis is underscored by the limited number of reported cases in the literature, as evidenced by the infrequent occurrence of this condition in newborns.

Description of the case. This case report presents a rare case of congenital epulis, a benign gingival tumor in a newborn. The report includes a brief literature review to provide insights into the clinical characteristics, diagnostic approaches, and management of congenital epulis.

Conclusion. This case contributes to the current knowledge regarding rarity of occurrence and emphasizes the need for reporting, early intervention and multidisciplinary collaboration in effective management.

Keywords. alveolar ridge, congenital epulis, gingiva, granular cell tumor, newborn

Introduction

Neonatal congenital epulis (CE) is an uncommon tumor. It is referred to as granular cell tumor or congenital gingival granular cell tumor due to its unique histologic characteristics. The term congenital granular cell epulis is the one used by the WHO.[1] Newmann originally reported it in 1871, and its 9–10:1 ratio indicates a preference for females.[2-4] The anterior maxillary alveolar ridge is where epulis is mostly seen.[5] It typically manifests as a single mass, while 10% of cases involve multiple lesions. It can originate from the tongue or mandibular gingiva as well.[2,6] It typically stops growing after birth.[7]

Clinically, congenital epulis presents as a pedunculated protuberant lump that might obstruct breathing or food intake. One still does not know the precise histogenesis. Prompt surgical resection is the advised course

of action. There have been no instances of malignant transformation, tumor recurrences, or harm to the dentition in the future.[6] A small number of instances with congenital epulis have been observed to spontaneously resolve.[8]

Aim

We summarize the pertinent literature and report on a female neonate who has congenital epulis, a solitary tumor in the anterior mandibular alveolar ridge.

Description of the case

A 6-hour old female neonate was brought into the out born section of the Neonatal intensive care unit (of a medical facility in Benin City), via Accident and Emergency accompanied by the father with complaints of a 'big' growth noticed on the mandibular alveolar ridge immediately after birth.

Mother was a 28-year-old P3 + 0 with secondary level of education. Her pregnancy was not scheduled at any hospital. However, the mother went to a traditional birth attendant at intervals for 'checkups.' No history suggestive of preeclampsia or bleeding per vagina in the antenatal period. There is history suggestive of malaria in the third trimester of pregnancy for which some herbal concoctions (contents unknown) were given to her by the trained afro-traditional birth attendant. The child was born at term via spontaneous vaginal delivery at the traditional birth attendant maternity home after about 12 hours of labor. No history of premature rupture of membrane or peripartum pyrexia.

The neonate had an APGAR score of 8/1, 9/5 and the birth weight was 2.7 kg. After birth, no bleeding or discharge was noticed from the growth which was large enough to be apparent. Growth was single and no other similar lesions were observed on other parts of the body.

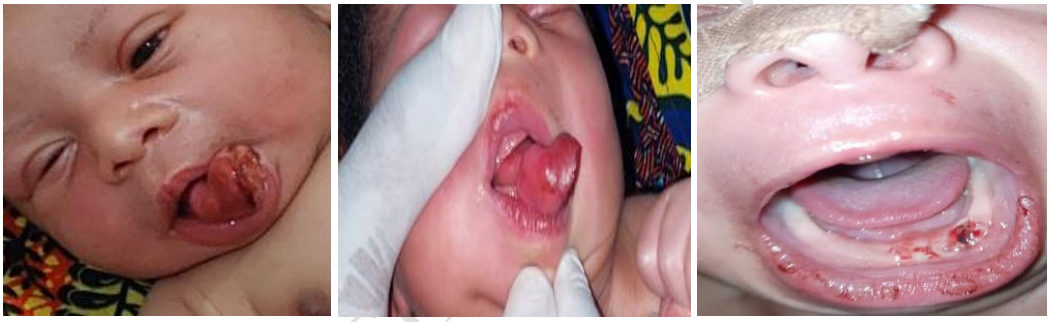
On presentation, child was afebrile, anicteric, acyanosed, not dehydrated, not pale, and not in any painful or respiratory distress. Random blood sugar was 48 mg/dL as the baby had not been fed after birth. Hypoglycemia was promptly corrected as per protocol. Vital signs were as follows: temperature: 36.4°C, heart rate: 128 bpm, respiratory rate: 44 cpm, SpO_2 in room air: 99%. On examination, a 3×2 cm ovoid pedunculated mass was noticed on the left aspect of the mandibular alveolar ridge (Fig. 1). The mass was non-tender, pink in color, fluctuant, and vascular. No spinal or bony deformities were observed. The systemic examination done was insignificant. A clinical diagnosis of congenital epulis was made. Differential diagnosis of granular cell tumor, hemangioma, rhabdomyosarcoma, dermoid cyst, fibroma and teratoma were made.

The neonate was placed on appropriate intravenous fluid and samples were also taken for full blood count and electrolyte, urea and creatinine which were within normal values. The pediatric surgeon and the maxillofacial surgeon were promptly informed to review.

On the first day of life, fluid was changed from 10% dextrose water to 8% dextrose saline. Upon review of the neonate by the pediatric surgeon, a nasogastric tube was passed for feeding with infant formula starting at 15 mL two hourly. No feeding difficulties or regurgitation was noticed. It was also agreed that the tumor was resectable and that it will be carried out by a maxillofacial surgeon. Daily cleaning of the growth two hourly with normal saline after yellowish slough was noticed was also commenced.

Due to hospital administrative issues and without affecting the patient's life, the surgery was delayed. During the successive days, vital signs were stable, and volume of feeds were increased by 5–10 mL every day which was well tolerated by the neonate. On the 8th day of life, the decision for surgery was made after payment and all necessary logistics had been carried out. At about 12 midnight, feeding was stopped, and baby was placed on nil per oral. Surgery was done the next day under general anesthesia. An incision was made to the peduncle and minimal bleeding was controlled with diathermy. The growth was excised and sent to the laboratory for histology.

Feeding was recommenced at 11 PM that night. Baby was placed on intravenous ceftazidime and genticin for five days duration, paracetamol for three days and was also breastfed from two days post operation till discharge on the 12th day of life. The neonate was discharged in a stable condition and was scheduled for a follow up visit in a week.



A **B** **C**

Fig. 1. A and B: Congenital epulis with a lengthy base linked to the lower bridge of the gingiva, C: Post-operation wound

Histopathological examination showed a polypoidal fragment of tissue lined by stratified squamous epithelium with a thin layer of parakeratosis. The sub-epithelial stroma is composed of nodules of large polygonal cells with eosinophilic granular cytoplasm and small central nuclei. There is no focus of atypia. Features were consistent with that of congenital granular cell tumor (Fig. 2). The tumor was also reckoned to be benign and capable of spontaneous regression.

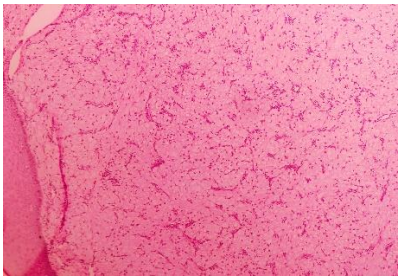


Fig. 2. Hematoxylin and eosin stain × 100 showing stratified squamous epithelium with a thin layer of parakeratosis (H&E, 100×)

Discussion

The etiology of congenital gingival granular cell tumor is unknown. It is believed to emanate from primitive neural crest mesenchymal cells. Histopathologic examination is required for definitive diagnosis.[9]

Prenatal diagnosis has made it possible to institute a multidisciplinary approach to its management involving the neonatologist, ear, nose, and throat surgeon, pediatric surgeon, and pediatric anesthetist. A magnetic resonance imaging (MRI) of the fetal brain could prove useful as it will outline the characteristics of the mass.[10] It would show a mixed mass with fibrofatty, mucosal and cartilaginous components.[11]

This neonate had no prenatal investigations done since the parents registered with a traditional birth attendant instead of registering at a hospital with proper antenatal care. Ultrasound has been shown to be effective in visualizing this lesion as early as 26 weeks gestation.[12]

Prenatal ultrasound or MRI may show a very large mass that would make vaginal delivery difficult, and a caesarian section can be planned for. Prenatal diagnosis can therefore be useful in instituting counselling and discussing the modality of intervention with parents and caregivers [1,13,14]

Due to its appearance, its occurrence can be frightening to the parents and caregivers of the neonate hence a good knowledge of this rare but benign tumor is necessary. Very few cases are reported in literature and to the knowledge of the authors this rare condition has only been reported once in Benin City by Edetanlen and Ogboh, 2019.[15]

The tumor commonly occurs at the maxillary alveolar ridge but in this present patient, it was found in the mandible.[16] However, surgical excision is generally indicated due to interference with feeding or respiration.[17]

The differential diagnosis of congenital epulis include granular cell tumor. Granular cell tumor though similar histologically to congenital epulis unlike it rarely affects neonates, is rarely located at the gingiva, and shows reactivity to S-100 protein and laminin.[18]

Historically, the anomaly has traditionally been treated with either broad or specific anesthesia and total removal via surgery. A case study including the use of a CO_2 laser with general anesthesia has also been published.[19] Congenital epulis has not been known to develop into cancer or grow after delivery. There

have been no reports of recurrence, even with partial excision. Incomplete resection has not been shown to cause a recurrence and most lesions have a penchant to involute.[8] There have been a few documented cases of complete regression without surgical intervention. [1,16]

It is a rare tumor with few case reports. A review of cases from 2000 to 2017 by Sohal et al. reported 156 cases globally.[20] This report was intended to add to the body of knowledge of the few cases reported so far from Nigeria and other African countries.[21-25]

Conclusion

Congenital epulis is a very rare benign lesion that affects the oral cavities of neonates. The small number of instances that have been described in the literature clearly indicates how uncommon this ailment is. It can sometimes cause problems with breathing and eating due to its large size, so it's important to make a diagnosis as soon as possible and the right care instituted. Early surgical intervention is important because deglutition and respiratory difficulty can be severe though spontaneous regression without intervention has been documented in the literature. An analysis of clinical as well as histopathological data is key to making an accurate diagnosis of this rare lesion.

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Declarations

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Author contributions

Conceptualization, S.O.,O.A.E. and K.A.; Methodology, S.O. and O.A.E.; Formal Analysis, S.O.,O.A.E. and K.A; Investigation, S.O.,O.A.E. and K.A; Resources, S.O.,O.A.E. and K.A; Data Curation, K.A.; Writing – Original Draft Preparation, S.O., and O.A.E.; Writing – Review & Editing, S.O. and K.A.; Supervision, K.A

Conflicts of interest

The authors declare no competing interests.

Data availability

No datasets were generated or analyzed during the current study.

Ethics approval

Before obtaining the report, the child's parents gave their informed permission.

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