

Congenital Epulis: A Case Report

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Abstract

Congenital epulis of newborn is a very rare benign soft tissue tumor of oral cavity. It occurs as a single mass with various sizes although some multiple lesions have also been reported. A large lesion may lead to respiratory distress and feeding difficulty or both. So it needs multidisciplinary team approach at the time of birth. Antenatal USG, perinatal MRI are adjunct to treatment planning particularly in large lesions. Though spontaneous regression in some cases have been reported. Treatment of choice is surgical excision. Recurrence after surgical removal is rare. We report a case of congenital epulis in a neonate presenting with feeding difficulty and was managed successfully with surgical removal. Histopathology of the excised lesion is important for final diagnosis.

Key words: congenital epulis, granular cell tumour of the newborn, Neumann's tumour, gingival tumour, newborn.

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I. Introduction

The term epulis is used to describe various gingival lesions and is derived from a Greek word meaning "of the gums". Congenital epulis, also known as congenital granular cell tumour (CGCT), granular cell fibroblastoma, congenital granular cell myoblastoma, or Neumann's tumour, is a very rare intraoral benign congenital soft tissue tumour. Neumann first described this case in the year 1871. And the reported incidence is about 0.0006%¹. The preferred and accepted term is congenital granular cell epulis of the newborn, which points to its non-neoplastic origin. This kind of epulis is a benign, reactive or degenerative lesion, usually single and typically situated on the alveolar mucosa of the jaws, most commonly on the maxillary alveolar ridge than the mandibular ridge (3 : 1). In 10% cases it may be located on both maxilla and mandible. Multiple lesions may be associated with other congenital anomalies like polydactyly, neuro fibroma, congenital goiter, nasal bridges^{2,3}. It is more commonly seen in female babies and has a 8-10 : 1 female to male ratio. This may indicate the role of a hormone in its development⁴. The aetio pathogenesis of the Neumann's tumour is still unclear. The antenatal diagnosis of epulis has been recently discussed in the medical literature. A large tumour can be diagnosed antenatally, particularly when associated with polyhydramnios⁵. Depending on location and size it may remain asymptomatic or symptomatic. In symptomatic cases the treatment of choice is surgical excision. Histopathology is confirmatory. Here in we share our experience in managing a case of congenital epulis in a newborn.

II. Case report

A hospital born, female baby, weighing 3 kg was shifted to NICU with an intra oral lesion, which was a smooth, sessile mass on the anterior ridge of the maxilla of size 2x1.5 cm at incisor site. The prenatal ultrasound showed no abnormalities. No family history of hereditary diseases was reported. The baby was born to a primi mother at 40th week of gestation with uneventful antenatal and perinatal history. The baby had no other associated congenital anomalies. There was feeding difficulty and was unable to suck breast milk properly. She had no respiratory distress. The lesion was well defined, firm, round, smooth and pink coloured. On day 3 of life the mass was excised after proper investigations, under general anaesthesia. The recovery was uneventful. She was discharged after 24 hrs. The microscopic examination of the lesions revealed nodular formations composed of homogeneous, large, round to oval histiocyte-like cells with distinct cell borders and abundant granular, eosinophilic cytoplasm, eccentrically placed, small, round, dark nuclei and absent nucleoli. The lesions were covered with normal squamous epithelium. The diagnosis was confirmed with histopathology. She was on regular follow ups for about one year and was doing well with no recurrence.



Fig. 1

Fig.2

Fig.3

**Fig.1 preoperative epulis
Fig. 2 intraoperative epulis
Fig.3.postoperative epulis**

III. Discussion

Congenital epulis more recently renamed by WHO as gingival granular cell tumour(GGCT) .It is a very rare lesion and arises from the mucosa of the gingiva, typically from the anterior part of the maxillary alveolar ridge, but may also arise from mandibular ridge (with the 3 : 1 maxillary predilection)^{6,7,8,9,10} . It is benign and usually a single mass, but can be multiple in 10% of the cases. Congenital epulis affects mostly white children and female to male ratio is 8-10 : 1¹ . The endogenous hormonal factors might influence growth in utero⁴ .

Congenital epulis usually is not associated with abnormality of teeth or other congenital malformations, the only exceptions are large lesions which can cause hypoplasia of the jaws, a hypoplastic or absent underlying tooth and the possibility of mild midface hypoplasia, known as Binder syndrome^{6,11} . The tumoral mass protruding through the mouth of a neonate can interfere with respiration and feeding, depending on the size¹⁰ . The pathogenesis is uncertain, however it is proposed to be originating from odontogenic epithelium, undifferentiated mesenchymal cells, pericytes, fibroblasts, smooth muscle cells, nerve related cells, and histiocytes^{7,8} . The currently accepted theory is that the epulis represents a reactive entity¹¹ . Immunohistochemical staining and ultrastructural examination favour myofibroblasts as the cell of origin.

Depending on size and location they may be symptomatic with respiratory or feeding difficulties or may remain asymptomatic. The diagnosis is always clinical and is confirmed with histopathology¹.

Our case had a single sessile lesion without any abnormality of teeth or associated congenital anomalies and had feeding difficulty. There was no respiratory distress.

The main differential diagnosis for the congenital epulis is adult granular cell tumour (myoblastoma). Both congenital and adult GCT have histopathologically identical cells. Early onset, unique location, pedunculated appearance, absence of pseudoepitheliomatous hyperplasia of the overlying squamous mucosa and no nerve bundles make the epulis easily differentiated from myoblastoma^{7,11,12}. It should also be differentiated from fibroma, rhabdomyoma, granuloma, Epstein's pearls, haemangioma, encephalocele, dermoid cyst, teratoma, melanocytic or pigmented neuroectodermal tumours, lymphatic malformations, schwannoma and heterotopic gastrointestinal cyst^{7,13}.

A very large lesion can be diagnosed in antenatal period with ultra sonogram as early as 26 wks which helps in planning the mode of delivery, but the findings are not specific^{7,12}. In the cases of patients with large lesions, polyhydramnion may be observed due to obstruction of oral cavity. In large lesions post natal MRI may aid assessment of the extent of lesion. In our case, the ultrasound performed in the first, second and third trimester of pregnancy did not reveal any abnormality. As the tumor was of not very large size we did not opt for MRI. Surgical excision is curative in symptomatic cases. There are no reports of recurrence or malignant transformations following surgery, even after incomplete removal¹¹. Very few cases of spontaneous regression of small lesions have also been reported^{1,14}. Asymptomatic cases may be observed for spontaneous regression. Radical excision is avoided as there is chance of damaging developing tooth buds^{1,2}

Histopathology is always confirmatory. As the baby had feeding difficulty the mass was excised surgically and the diagnosis was made basing on histopathology report.

She was followed up to one year of age and was doing well. No recurrence or teeth anomaly were detected.

IV. Conclusion

The congenital epulis is a rare lesion of the oral cavity present at birth. The tumor is often misdiagnosed because of its rarity. It may be quite distressing to parents. Early diagnosis and proper treatment is necessary in symptomatic cases.

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