

Multiple Congenital Epulis in Newborn: A Case Report

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Abstract

Case Report

Congenital Granular epulis is a rare soft tissue lesion of the newborn. It presents as a benign mass lesion in oral cavity, most commonly arising from the alveolar processes. It is composed of sheets and nests of cells with abundant granular cytoplasm [1]. Here, we report a case of congenital epulis in a seven day old newborn female child, which were multiple, and present on the right upper and right lower gingiva since birth.

Keywords: Epulis, congenital granular cell tumor, Neumann's tumor.

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INTRODUCTION

According to world health organization (WHO) classification, congenital gingival granular cell epulis (CGCE) is also known as congenital epulis, congenital epulis of the newborn, congenital gingival granular cell tumor, Neumann's tumor [2]. Congenital granular cell epulis affects newborn. Most cases commonly showing female preponderance [3]. Most cases develop on the maxillary anterior alveolar process, although it can occur on the mandibular process also [4]. Clinically, Congenital granular cell epulis predominantly presents as a smooth surfaced sessile or pedunculated soft tissue mass of normal or reddish colour mucosa, with varying size ranging from <1 cm to several centimetres in diameter [2-5]. Hence, this tumor due to varying size may cause obstruction in respiration and feeding [6]. Congenital epulis is although a rare tumor of newborn, the presence of multiple masses over various sites is still a rarer, having been reported in approximately 10% of cases [7]. Thereby, we report a rare case of congenital granular cell epulis, with multiple masses describing the clinical, radiological, histopathological and immunohistochemical profile.

CASE REPORT

A seven day old newborn female child presented to our hospital with swelling in the right lower jaw and right upper jaw since birth. The baby also complained of fever with chills and cough with yellowish coloured sputum since birth. There was

difficulty in feeding. There was no history of trauma. No significant past or family history was noted.

On examination, the general condition was moderate with stable vitals. On local examination, a round soft mass of size 1.3x1.5cm was located on right lower jaw, and two swellings of size 0.5x0.5cm over right upper jaw. All the laboratory tests were within normal limit.

Imaging findings: A CT Brain (Plain+contrast) was performed and axial sections of the brain were obtained. The CT Brain (Plain+contrast) revealed a well defined smooth walled homogeneously enhancing soft tissue attenuation lesion measuring 11x16x10 mm (APXTRXCC), seen in the right paramedian lower jaw region most likely arising from the alveolus (Figure-1). The CT impression was given as right lower jaw benign neoplastic mass, possibly congenital epulis; requiring histopathological confirmation.

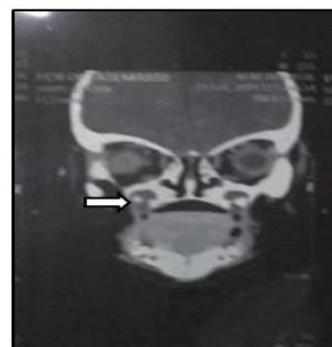


Fig-1: CT Brain (Plain+contrast)

The tumor was completely resected by surgical excision following anaesthesia. The intra operative findings were as follow; a 2x2x0.5 cm soft swelling arising from right lower alveolus and two tiny swellings approximately 4x3 mm were seen arising from right upper alveolus with no bony involvement. Post operative period was uneventful and newborn was tolerating feeds well.

The specimens were fixed in 10% formalin and sent for histopathological examination. We received masses in two containers. Container one was labelled as 'right lower alveolus mass' containing a single, white, firm, well circumscribed, slightly lobulated mass measuring 1.5x1x0.5 cm. The cut surface was white, homogenous, firm. The second container was labelled as 'right upper alveolus mass' containing two grey white tissue bits measuring 0.5x0.2x0.1cm respectively (Figure 2 & 3).

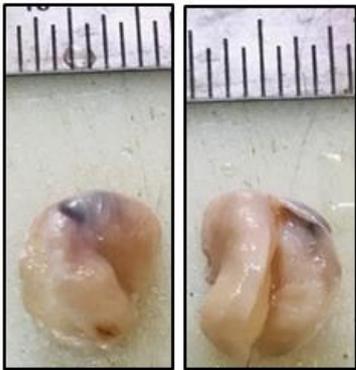


Fig-2: Two Right upper alveolar masses

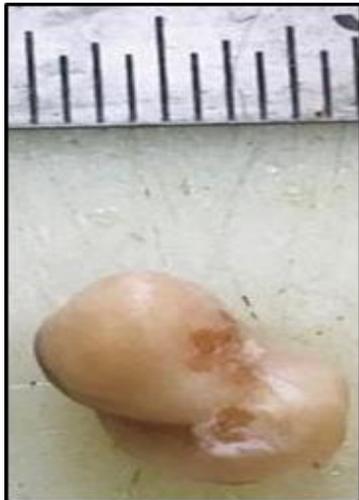


Fig-3: Single right lower alveolar mass

Microscopic examination of the hematoxylin and eosin (H&E) sections from both the masses revealed similar histological features. Sections showed a benign nodular lesion focally covered by stratified squamous epithelium (Fig-4). The underlying sub-epithelium showed a tumor composed of sheets of polygonal to oval cells with small vesicular nuclei and abundant granular eosinophilic cytoplasm. The tumor

showed increased vascularity (Fig 5 & 6). The histologic findings from both the masses were consistent with the diagnosis of congenital granular cell epulis. There was no evidence of atypia or malignancy in the sections studied.

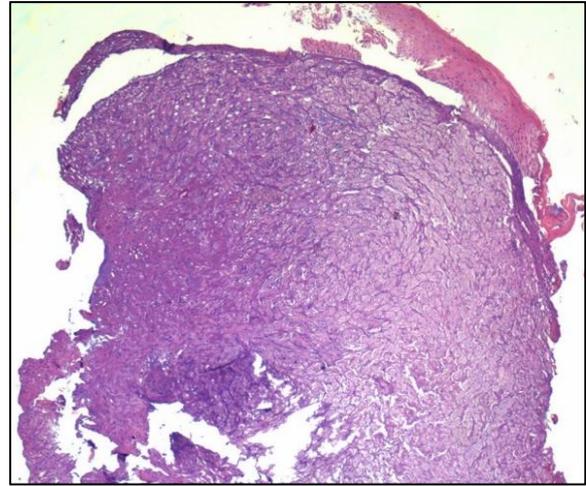


Fig-4: H&E stain, x10

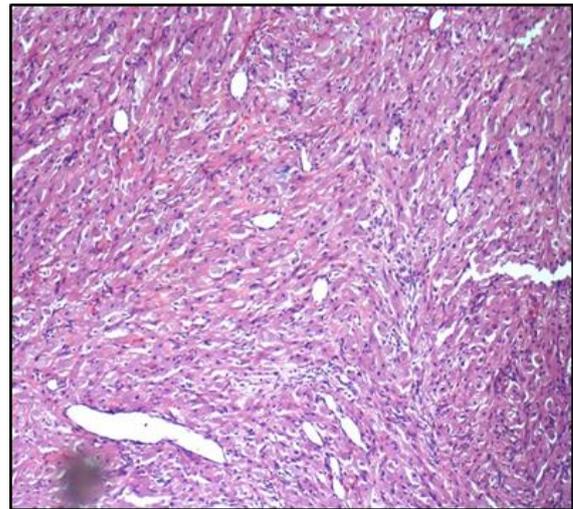


Fig-5: H&E stain, x40

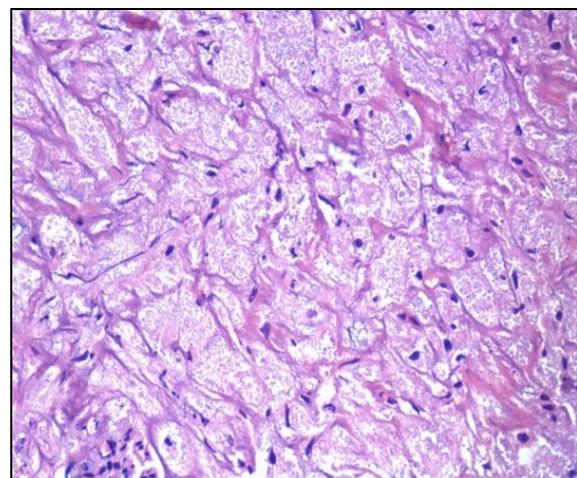


Fig-6: H&E stain, x100

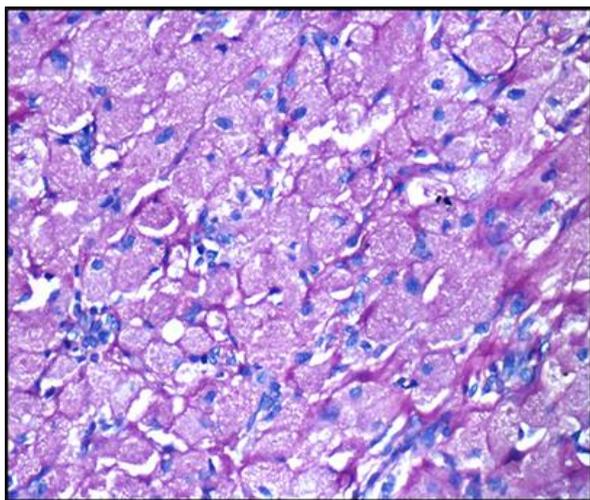


Fig-7: PAS stain, x 100

Special stain and Immunohistochemical finding: Positive staining was found for Periodic Acid Schiff stain, a glycogen marker (Fig-7). S-100 stain was negative. Hence, these findings were consistent with congenital granular cell epulis.

DISCUSSION

This benign tumor which appears rarely has different names such as congenital epulis (CE), congenital gingival granular cell tumor of the newborn, or Neumann's tumor [2].

Usually, it appears as a single lesion, but in 10% cases it may arise from multiple locations simultaneously [8]. Thus in our case, the lesion occurred in multiple locations along right lower jaw and upper jaw also. The size of the tumor may vary from a few millimeters to several centimeters in diameter. Due to the difficulty in swallowing and postnatal feeding, due to difficulties in breathing, multiple lesions may cause severe problems in neonate. Our case of a Congenital epulis was a female newborn diagnosed with three pedunculated masses, single soft swelling arising from right lower alveolus and two tiny swellings were seen arising from right upper alveolus, which caused difficulty in feeding.

The most common site of the origin is maxillary alveolar process. Other sites are tongue, palate, skin, subcutaneous tissue, skeletal muscles. About 90% cases are seen in female neonates with a female: male ratio of 8:1. It has no tendency of local recurrence or malignant transformation [9].

Histologically, congenital epulis is similar to the adult granular cell tumor. They are both composed of large round cells with abundant granular cytoplasm and small eccentric nuclei with occasional small nucleoli. There is a prominent vascular stroma with perivascular lymphocytes and histiocytes. In contrast to adult granular cell tumors, there is no pseudoepitheliomatous hyperplasia of the overlying

squamous mucosa in congenital epulis, and no nerve bundles are seen within this lesion. Furthermore, congenital epulis was shown to be consistently negative when immunostained with S-100 protein, in contrast to adult granular cell tumors, which are derived from Schwann cells [10]. Similarly, in our case the tumor was negative for S-100 protein, thus confirming the diagnosis of Congenital epulis.

The gross differential diagnosis of oral cavity lesions in the newborn includes hemangioma, lymphangioma, teratoma, pigmented neurectodermal tumor of infancy, rhabdomyoma and rhabdomyosarcoma [11].

Because of the multiple locations, increased size of the tumor, and risk of interference with breathing and feeding, complete surgical excision is the treatment of choice. Recurrence rarely occurs, and the prognosis is reported to be excellent [12]. Spontaneous regression has been reported in isolated few cases and may occur even in case of incomplete resection [13].

Even though it is a benign lesion, but due to multiple locations it may obstruct the oral passage by its size and induce breathing and feeding problems. Delay in operation may cause airway obstruction and feeding difficulty. The tumor should be removed during the immediate postnatal period, which was done in our case.

CONCLUSIONS

Congenital granular cell epulis is a benign tumor occurring predominantly in the alveolar maxillary ridges of newborn, with the occurrence of multiple locations is rare. The multiple lesions are of concern, since it may cause interference in feeding and breathing. Hence, clinical follow-up with accurate diagnosis and appropriate treatment is necessary in such conditions.

Conflict of Interests: The authors declare that there is no conflict of interests regarding the publication of this paper.

REFERENCES

1. Conrad R, Perez MC. Congenital granular cell epulis. *Archives of Pathology and Laboratory Medicine*. 2014 Jan;138(1):128-31.
2. Van der Waal I, Barnes L, Eveson J. W, Reichart P, Sidransky D, editors. *Congenital granular cell epulis*. World Health Organization Classification of Tumours. Pathology and Genetics. Head and Neck Tumours. Lyon, France: IARC Press; 2005;198.
3. Yuwanati M, Mhaske S, Mhaske A. Congenital granular cell tumor—a rare entity. *Journal of neonatal surgery*. 2015 Apr;4(2):17..

4. Childers EL, Fanburg-Smith JC. Congenital epulis of the newborn: 10 new cases of a rare oral tumor. *Annals of diagnostic pathology*. 2011 Jun 1;15(3):157-61.
5. Kumar R, Jaiswal S, Singhal A, Garg R. Congenital granular cell lesion: A rare tumor of new born. *Journal of oral and maxillofacial pathology: JOMFP*. 2013 Sep;17(3):440-442.
6. Küpers AM, Andriessen P, van Kempen MJ, van der Tol IG, Baart JA, Dumans AG, van der Waal I. Congenital epulis of the jaw: a series of five cases and review of literature. *Pediatric surgery international*. 2009 Feb 1;25(2):207-10.
7. Dzieniecka M, Komorowska A, Grzelak-Krzymianowska A, Kulig A. Multiple congenital epuli (congenital granular cell tumours) in the newborn: a case report and review of literature. *Polish Journal of Pathology*. 2011;62(1):69-71.
8. Karthiga SK, Rajesh R. Congenital epulis—congenital granular cell lesion: a case report. *Journal of Indian Society of Pedodontics and Preventive Dentistry*. 2006;24(2):104-106.
9. Wittebole, A, Bayet, B, Veyckemans F, Gosseye S, Vanwijck R. Congenital epulis of the newborn. *Acta ChirBelg*. 2003;103:235-237.
10. Lack EE, Worsham GF, Callihan MD, Crawford BE, Vawter GF. Gingival granula cell tumors of the newborn (congenital" epulis"): a clinical and pathologic study of 21 patients. *The American journal of surgical pathology*. 1981 Jan;5(1):37-46.
11. Hasanov A, Musayev J, Onal B, Rahimov C, Farzaliyev I. Gingival granular cell tumor of the newborn: a case report and review of literature. *Turk Patoloji Derg*. 2011 May 1;27(2):161-3.
12. Bang, K, Bodhade A, Dive A. Congenital granular cell epulis of a newborn. *Dent Res Journal*. 2012;9:136-8.
13. Sakai VT, Oliveira TM, Silva TC, Moretti AB, Santos CF, Machado MA. Complete spontaneous regression of congenital epulis in a baby by 8 months of age. *International journal of paediatric dentistry*. 2007 Jul;17(4):309-12.