

CONGENITAL EPULIS: A CASE REPORT

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ABSTRACT

Congenital epulis is a rare lesion of the newborn. A case of congenital epulis is presented here with details on the clinical, histopathological characteristics as well as the management of the lesion.

Key words: *Congenital epulis, anterior maxillary alveolar ridge, female infants*

INTRODUCTION

The congenital epulis of the newborn is an uncommon benign soft tissue tumor that occurs on the alveolar ridge of the newborn. It appears predominantly on the anterior maxillary alveolar ridge.¹ However rare cases of congenital epulis have been reported on the tongue.² Some pathologists prefer using the term congenital granular cell lesions, since not all cases are presented as an epulis on the alveolar ridge.

The first case was reported by Newmann in 1871¹, since then more than 200 cases have been documented in the English language literature,³ with a female predilection, 10 times more often than in males.¹ Congenital epulis usually occurs as a solitary lesion, however multiple lesions involving maxilla, mandible and or the tongue have also been reported.^{4,5} This lesion is usually treated by surgical excision and no recurrence has been so far reported.⁶

The purpose of this article is to present the clinical and histopathological features of congenital epulis in a new born female reported at the College of Dentistry, King Saud University, Riyadh, Saudi Arabia.

CASE REPORT

A one month old female infant was referred to the oral and maxillofacial surgery department at the College of Dentistry, King Saud University, for evaluation and treatment of a soft tissue mass protruding from the oral cavity. Intraoral examination revealed pink, polypoid soft tissue mass with smooth surface, approxi-

mately 2 cm in diameter, attached to the anterior of the right maxillary alveolar ridge. The lesion did not interfere with feeding or respiration, but prevented complete closure of lips (Figure 1). Because of the classical clinical features of the lesion, a preliminary diagnosis of congenital epulis was considered. The tumor was excised under general anesthesia. The postoperative course was uneventful.

The specimen was accordingly processed for light microscopic examination. The excised lesion was fixed in 10% neutral buffered formalin, followed by paraffin embedding. Histological sections from the paraffin-embedded blocks were stained with hematoxylin and eosin.

The microscopic examination revealed a tumor composed of rounded and polygonal cells with abundant granular eosinophilic cytoplasm and round to oval, lightly basophilic nuclei. Scattered blood capillaries were noted throughout the lesion. The tumor was covered by parakeratinized stratified squamous epithelium with normal architecture (Figures 2, 3 & 4). Based on the clinical and the histopathological findings, a definitive diagnosis of congenital epulis of the newborn was established.

DISCUSSION

Congenital epulis is a benign intraoral tumor seen in infants. This tumor typically appears as erythematous lobulated mass on the alveolar ridge of the newborn. Most examples are 2 cm or less in diameter,⁶ although lesions as large as nine centimeter in diam-

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Fig 1: Congenital epulis is protruding from the oral cavity.

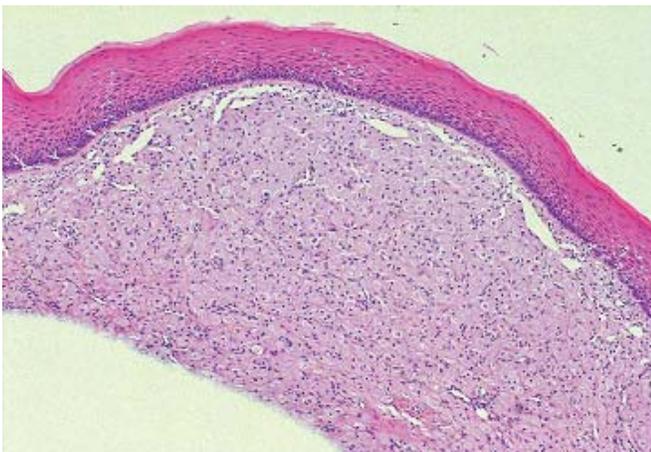


Fig 2: H&E stain (original magnification x 100) showing tumor cells of congenital epulis surfaced by parakeratinized stratified squamous epithelium with smooth basement membrane.

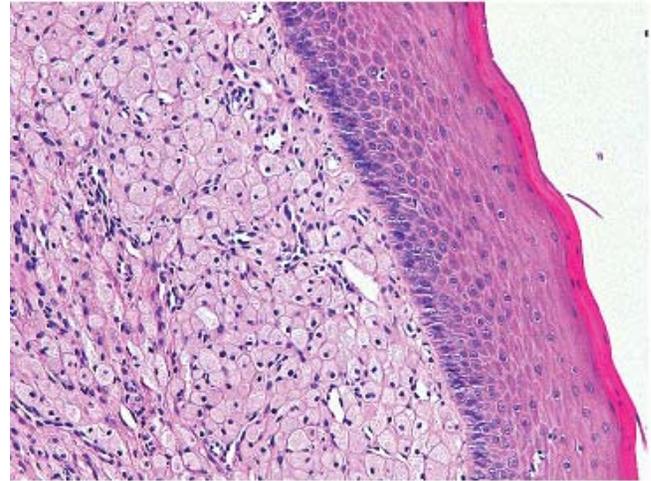


Fig 3: H&E stain (original magnification x 200) showing congenital epulis consisting of rounded and polygonal cells with abundant granular eosinophilic cytoplasm, Scattered blood capillaries are noted throughout the lesion

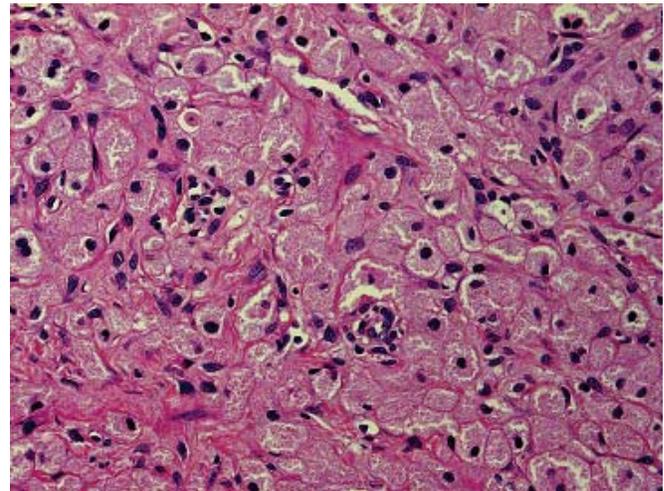


Fig 4: H&E stain (original magnification x 400) showing rounded and polygonal cells with abundant granular eosinophilic cytoplasm and round to oval lightly basophilic nuclei.

eter have also been reported.⁷ The lesion occurs on the maxillary alveolar ridge twice as often as on the mandible. It frequently arises lateral to the midline in the area of the developing lateral incisor and canine teeth.^{1,6}

Congenital epulis shows a striking predilection for females. Females are affected 10 times more often than males,¹ which may suggest a hormonal influence during its course of development,⁸ however estrogen and progesterone receptors have not been detected.⁹

Congenital epulis usually occurs as a solitary nodule on the anterior alveolar ridge of either jaw; however multiple tumors are seen occasionally. In 10% of cases multiple tumors have been noted on the same or different alveolar ridges,^{1,10} in addition few rare examples on the tongue have been described in infants who also had simultaneous occurrence of alveolar tumors.^{4,5,11}

Although the term “congenital epulis” is commonly used by oral pathologist to describe this lesion, several other diagnostic terminologies have also been used;

such as congenital granular cell myoblastoma^{12,13} and gingival granular cell tumor⁶ because of the microscopic resemblance of this lesion to the granular cell tumor. These terms are inappropriate (and their use should be avoided) because this lesion (congenital epulis) exhibits ultrastructural and immunohistochemical differences that warrant its classification as a distinct and separate entity.^{9, 14, 15} The term congenital epulis is commonly used by pathologists, however, in the recent World Health Organization classification of the head and neck tumors, this lesion has been named as “congenital granular cell epulis”.²

Histogenesis of congenital epulis is still uncertain, some recent studies lend support to its non-neoplastic nature.¹⁶ However, earlier investigations suggested that the origin of this lesion is from the undifferentiated mesenchymal cells based on their ultrastructural and immunohistochemical findings.^{9,14-17} The transmission electron microscopic investigations revealed tumor cells that were filled with autophagosomes, in addition occasional tumor cells demonstrated long dendritic processes that contained contractile microfilaments⁹. Immunohistochemical investigations of congenital epulis demonstrated negative S-100 protein staining and positive staining for vimentin.^{14,16,17} These observations suggest that the tumor cells may represent early mesodermal cells with myofibroblastic differentiation. In addition, these findings confirmed that this lesion has a different mesenchymal origin from granular cell tumor.^{9, 14-17}

Histologically, congenital epulis consist of round cells with granular eosinophilic cytoplasm and round to oval basophilic nuclei within vascular fibrous connective tissue. Contrary to the granular cell tumors, the overlying epithelium in this lesion never shows pseudoepitheliomatous hyperplasia but, typically exhibits a smooth epithelial – connective tissue interface.¹

Occasionally, the tumor has been detected in utero during ultrasound examination.^{3,18, 19} Recently, there has been an increased number of congenital epulis cases diagnosed through prenatal ultrasound diagnostic imaging, this further aid in the successful postnatal management and in turn reduce the complications associated with congenital epulis.^{3,18, 19}

This lesion is usually treated by simple surgical excision. No recurrence has been so far reported even

without its complete removal.^{1, 6} Eventual regression have been also noticed in few patients without undertaking any treatment,⁶ which further supports the concept of non-neoplastic reactive nature of this lesion.

SUMMARY

Congenital epulis is a rare lesion of the newborn. Understanding the clinical and histopathological features of this lesion are important in the successful outcome of its management.

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REFERENCES

- 1 Zuker RM, Buenechea R. Congenital epulis: review of the literature and case report. *J Oral Maxillofac Surg* 1993; 51:1040-1043.
- 2 Senoo H, Iida S, Kishino M, Namba N, Aikawa T, Kogo M. Solitary congenital granular cell lesion of the tongue. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2007; 104:e45-48.
- 3 Kim YD, Kim HJ, Lee NK, Ha WH, Lee CH, Park SE. Congenital epulis: prenatal ultrasonographic and postnatal MR features with pathologic correlation. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2008; 106:743-748.
- 4 Nouri S, Mahdhaoui N, Bouafsoun C, Zakhama R, Omezzine M, Tahar Yaacoubi M, et al. Obstructive congenital multiple epulis: A case report in a male neonate. *Arch Pediatr* 2008; 15:1083-1086.
- 5 Loyola AM, Gatti AF, Pinto DS, Jr., Mesquita RA. Alveolar and extra-alveolar granular cell lesions of the newborn: report of a case and review of literature. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 1997; 84:668-671.
- 6 Lack EE, Worsham GF, Callihan MD, Crawford BE, Vawter GF. Gingival granular cell tumors of the newborn (congenital “epulis”): a clinical and pathologic study of 21 patients. *Am J Surg Pathol* 1981; 5:37-46.
- 7 Chami RG, Wang HS. Large congenital epulis of newborn. *J Pediatr Surg* 1986; 21:929-930.
- 8 Roy S, Sinsky A, Williams B, Desilets V, Patenaude YG. Congenital epulis: prenatal imaging with MRI and ultrasound. *Pediatr Radiol* 2003; 33:800-803.
- 9 Damm DD, Cibull ML, Geissler RH, Neville BW, Bowden CM, Lehmann JE. Investigation into the histogenesis of congenital epulis of the newborn. *Oral Surg Oral Med Oral Pathol* 1993; 76:205-212.
- 10 Feller L, Wood NH, Singh AS, Raubenheimer EJ, Meyerov R, Lemmer J. Multiple congenital oral granular cell tumours in a newborn black female: a case report. *Cases J* 2008; 1:13.

- 11 Yavuzer R, Ataoglu O, Sari A. Multiple congenital epulis of the alveolar ridge and tongue. *Ann Plast Surg* 2001; 47:199-202.
- 12 Atterbury RA, Vazirani SJ. Granular-cell myoblastoma of the newborn; report of a case. *Oral Surg Oral Med Oral Pathol* 1957; 10:1037-1040.
- 13 Cussen LJ, MacMahon RA. Congenital granular-cell myoblastoma. *J Pediatr Surg* 1975; 10:249-253.
- 14 Lifshitz MS, Flotte TJ, Greco MA. Congenital granular cell epulis. Immunohistochemical and ultrastructural observations. *Cancer* 1984; 53:1845-1848.
- 15 Lack EE, Perez-Atayde AR, McGill TJ, Vawter GF. Gingival granular cell tumor of the newborn (congenital "epulis"): ultrastructural observations relating to histogenesis. *Hum Pathol* 1982; 13:686-689.
- 16 Vered M, Dobriyan A, Buchner A. Congenital granular cell epulis presents an immunohistochemical profile that distinguishes it from the granular cell tumor of the adult. *Virchows Arch* 2009; 454:303-310.
- 17 Mirchandani R, Sciubba JJ, Mir R. Granular cell lesions of the jaws and oral cavity: a clinicopathologic, immunohistochemical, and ultrastructural study. *J Oral Maxillofac Surg* 1989; 47:1248-1255.
- 18 McMahon MG, Mintz S. In utero diagnosis of a congenital gingival granular cell tumor and immediate postnatal surgical management. *J Oral Maxillofac Surg* 1994; 52:496-498.
- 19 Williams RW, Grave B, Stewart M, Heggie AA. Prenatal and postnatal management of congenital granular cell tumours: a case report. *Br J Oral Maxillofac Surg* 2009; 47:56-58.