J Pediatr Adolesc Surg. 2024; 2:54-56
DOI: https://doi.org/10.46831/jpas.v1i2.99

Case Report

Congenital epulis: Report of two cases

Ali Raza Chaudhry,[1*] Rumaisaa Saman,[1] Muhammad Umar Nisar,[1] Khawar Abbas,[2] Samer Sikander,[3]

1 Department of Paediatric Surgery, The Children Hospital, Pakistan Institute of Medical Sciences, Islamabad

2 Department of Paediatric Surgery, Shifa International Hospital, Islamabad

3 Paediatric Medicine, PAEC Hospital, Islamabad

Cite as: Chaudhry AR, Saman R, Nisar MU, Abbas K, Sikander S. J Pediatr Adolesc Surg. 2024; 2:54-56.

ABSTRACT

Background: Congenital epulis is a rare benign, smooth, sessile, or pedunculated, solitary mass of varying sizes commonly occurring at the anterior alveolar ridge of the maxilla. It can occur as a solitary as well as a multifocal lesion.

Case Presentation: Case 1: A two-day-old female newborn presented with a single round, smooth, pedunculated mass measuring 3x2x2cm on the anterior alveolar ridge of the maxilla. Complete excision of the mass was done under general anesthesia. Histopathology confirms congenital epulis. No recurrence was found till one year of follow-up. Case 2: A one-day-old female newborn presented with a 3x4x3cm soft to firm mass arising from the right upper alveolar margin attached with a stalk and extending into the mouth without any respiratory difficulty. Complete excision of the mass was done under general anesthesia. Follow-up for one year showed no recurrence.

Conclusion: Congenital epulis is a rare benign lesion most commonly arising from the anterior alveolar ridge of the maxilla without the involvement of bone or teeth. Complete surgical excision is the treatment of choice without any reported recurrence.

Keywords: Neumann's tumor, Granular cell tumor, Granular cell fibroblastoma, Congenital granular cell myoblastoma, Congenital gingival granular cell tumor.

INTRODUCTION

Congenital Epulis (CE), Neumans Tumor, Congenital Granular cell tumor (CGCT), Granular cell fibroblastoma, congenital granular cell myoblastoma, and congenital gingival granular cell tumor are different eponyms for this rare congenital benign tumor of a newborn. The first-ever case was reported in the literature by Neuman in 1871. The Greek term "Epulis" means "on the gingiva". As there are cases that are not exclusively related to the gingiva, so epulis is a non-specific but most commonly used term for these masses. However, congenital granular cell tumor seems more appropriate to describe these lesions. The tumor arises from the maxillary alveolar ridge, corresponding to the future location of the canine and lateral incisor, though other intraoral sites such as the mandible and tongue have also been reported. Clinically, the tumor presents as a smooth, sessile, or pedunculated, solitary mass of varying sizes from a few millimeters to as large as 9 cm, usually with normal overlying reddish mucosa. Females are mostly affected (8–10 females: 1 male) with a predisposition for the Caucasian race. [1, 2] Herein we report two cases of congenital Epulis.

CASE REPORT

Case 1

A two-day-old female newborn was admitted with a single rounded, smooth, pedunculated mass on the anterior alveolar ridge of the maxilla. No prenatal ultrasound was done. The neonate was born at term as simple vertex delivery in a peripheral hospital without any difficulty with a birth weight of 3650 g. The baby did not require any mechanical respiratory support. On local examination, a single fleshy pedunculated mass arose from an alveolar ridge measuring 3x2x2cm and to the right of the midline.

Correspondence*: Ali Raza Chaudhry, Department of Paediatric Surgery, The Children Hospital, Pakistan Institute Of Medical Sciences, Islamabad.
Email: alirchaudhry@gmail.com
Submitted on: 25-05-2021

Accepted on: 19-07-2021

Mass was well-circumscribed, firm, round, smooth, and pink in color (Fig. 1a). A nasogastric tube was passed for feeding and oxygen was attached as the baby was having respiratory difficulty. Excision of the mass was done under general anesthesia (Fig. 1b & 1c). Postoperative recovery was uneventful and the baby was discharged on the same day. The histopathology report showed a cluster of cells containing abundant granular eosinophilic cytoplasm with the presence of macrophages and lymphocytes, areas of hemorrhage, and ulceration. The histopathological findings were consistent with congenital epulis. Follow-up was done till one year of age with no sign of recurrence.

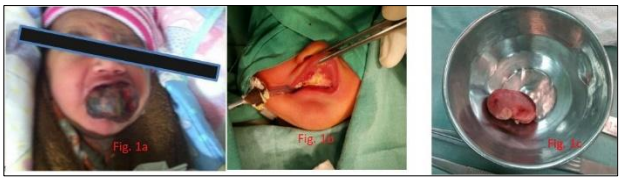

Figure 1a: Pre-operative picture showing rounded pedunculated mass arising from the oral cavity.
Figure 1b: Per-operative picture after excision of mass from the anterior alveolus.
Figure 1c: Excised mass

Case 2

A one-day-old female newborn was referred to us for a soft to firm swelling on the right upper gum with a free lower margin extending into the mouth. It was full-term pregnancy, simple vertex delivery with episiotomy and the baby was weighing 2600 g. The baby was shifted to NICU with a heart rate of 128/min and respiratory rate of 58/min. After careful examination, the patient was referred to our hospital with suspicion of teratoma. On examination, a soft swelling about 3x4 cm arising from the right upper alveolar margin of the maxilla and extending into the mouth was observed (Fig.2).

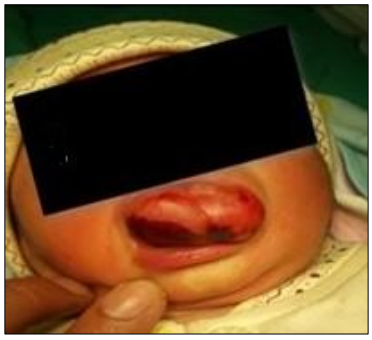

Figure 2: Pre-operative picture showing rounded mass arising from the oral cavity

The mass was excised in toto along with frenotomy of the upper lip under general anesthesia. Postoperative recovery was uneventful. The patient was discharged on the same day on full feeding. Histopathology report showed fragments showing stratified squamous epithelial covering, exhibiting focal hyperplasia. Underlying tissue showed proliferation of fibroblasts and few multinucleated giant cells consistent with epulis. Follow-up after one year showed no recurrence.

DISCUSSION

Congenital Epulis (CE) is a benign tumor of the newborn most commonly arising from the alveolar ridge of the maxilla as a pedunculated mass with smooth reddish mucosal covering. It can also occur in the alveolus of the mandible which is a less common location. Mostly it occurs as a solitary mass but sometimes multiple lesions [3]can also be found in 10% of cases leading to respiratory and feeding difficulty. Maxilla and mandible both are often involved in multifocal lesions.[4,5] Involvement of bones and teeth is rare. Our both cases had no involvement of bone or teeth.

CE has a prenatal growth pattern and it grows rapidly during the third trimester. CE doesn't grow after birth and shows occasional spontaneous regression as well. Prenatal diagnosis using ultrasonography can be made as early as 26 weeks of gestation as a well-defined hypoechogenic mass protruding out of the oral cavity and comprising of branching vascular pattern.[6]

Histologically CE appears as well-circumscribed nests and ribbons of homogenous to slightly spindle-shaped tightly packed cells having eosinophilic granular cytoplasm and eccentric nuclei without any mitosis. There is a prominence of capillaries with slight thinning of overlying stratified squamous epithelium. Variations in histologic appearance are also observed in an old and traumatized lesion, such as increased fibrosis and spindle cell proliferation. Immunologically, CE is positive for vimentin and neuron specific enolase. [6, 7]

It is believed that it develops reactively in response to maternal hormones during pregnancy. Tendency to decrease in size or even spontaneous resolution after birth as maternal hormones reduce, supports this theory. Additionally, no recurrence even after incomplete excision and lack of malignant transformation also support this theory. [8, 9]Differential diagnoses include adult granular cell tumor (GCT), fibroma, rhabdomyosarcoma, hemangioma, granuloma, cephalocele, Epstein pearls, teratoma, melanocytic neuroectodermal tumors, lymphatic malformations, heterotopic Gastro-intestinal cysts, congenital cystic choristoma, lipoma, and schwannoma. Of these, adult GCT is the main differential based on histopathology as both of these lesions have similar cells. Important differentiating points for epulis include earlier presentation, origin, gross appearance as a pedunculated lesion, lack of pseudo-epithelial hyperplasia of overlying squamous mucosa, and absence of nerve bundles.[8,9] Adult GCT mostly originates from the tongue, soft palate, and floor of the mouth.[4]

Treatment of CE is surgical. Complete excision is the treatment of choice in large lesions. However, radical excision should be avoided to prevent damage to the underlying bone and developing tooth buds as there are no reports of recurrence even after incomplete excision of the tumor. In very small tumors there are reports of spontaneous regression as well. Therefore in such lesions, continuous observation is also advocated.[10,11]

In conclusion, congenital epulis is a rare benign lesion most commonly arising from the anterior alveolar ridge of the maxilla without the involvement of bone or teeth. Complete surgical excision is the treatment of choice without any reported recurrence.

Conflict of Interest: Nil

Source of Supports: Nil

Consent to Publication: Author(s) declared taking informed written consent for the publication of clinical photographs/material (if any used), from the legal guardian of the patient with an understanding that every effort will be made to conceal the identity of the patient, however it cannot be guaranteed.

Authors Contribution: Author(s) declared to fulfill authorship criteria as devised by ICMJE and approved the final version. Authorship declaration form, submitted by the author(s), is available with the editorial office.

Acknowledgements: None

REFERENCES

1. Aparna H, Jayanth B, Shashidara R, Jaishankar P. Congenital epulis in a newborn: a case report, immunoprofiling and review of literature. Ethiopian J Heal Sci. 2014; 24:359-62.
2. Bhat C, Chaugule V, Patil V, Shah K. Congenital epulis in newborn: Case report. New York State Dental J. 2012; 78:50.
3. Iqbal A, Bhosale M, Soni V, Anand S. Multiple lesions of congenital epulis. J Neonatal Surg 2021(6);10:28.
4. Cheung JM, Putra J. Congenital granular cell epulis: Classic presentation and its differential diagnosis. Head Neck Pathol. 2020; 14:208-11.
5. Bang KO, Bodhade AS, Dive AM. Congenital granular cell epulis of a newborn. Dental Res J. 2012; 9:S136
6. Conrad R, Perez MCN. Congenital granular cell epulis. Arch Pathol Lab Med. 2014; 138:128-31.
7. Damante JH, de Souza TE, Mazzottini R, Monteiro-Amado F, Fleury RN, Soares CT. Congenital granular cell lesion: clinical, microscopic and immunohistochemical aspects in a case of multiple lesions. J Clin Pediatr Dent. 2011;36(1):71-4.
8. 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. Pol J Pathol. 2011;62(1):69-71.
9. 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;27(2):161-3.
10. Hiradfar M, Zabolinejad N, Gharavi M, Sebt S. Multiple congenital epulis of the mandibular ridge: a case report. Iranian journal of otorhinolaryngology. 2012;24(69):193.
11. Kato H, Nomura J, Matsumura Y, Yanase S, Nakanishi K, Tagawa T. A case of congenital granular cell epulis in the maxillary anterior ridge: a study of cell proliferation using immunohistological staining. J Maxillofac Oral Surg. 2013;12(3):333-7.