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Surgical Management of a Large Congenital Granular Cell Epulis Arising From the Anterior Maxillary Alveolar Ridge in a Newborn: A Case Report

Anusha Karre [1], Shaik Hasanuddin [1], K Suhasini [1], B Simren [1], Prasanna Miyapuram [1]

1. Pediatric and Preventive Dentistry, Government Dental College and Hospital, Hyderabad, IND

Corresponding author: Anusha Karre, anushakmds@gmail.com

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Abstract

Congenital epulides are rare benign developmental lesions believed to originate from primitive mesenchymal cells of the cranial ectomesenchyme, with their exact histogenesis remaining uncertain. These lesions may occasionally attain a considerable size, resulting in functional difficulties in newborns. Often diagnosed as congenital granular cell epulis, based on their histopathology and immunohistochemistry, these lesions are confused with congenital hemangiomas, teratomas, and odontomas. In the present case report, a 10-day-old female infant was presented with a large, soft, gingival outgrowth, which was provisionally diagnosed as a congenital granular cell epulis. Even though prenatal diagnostic tools are available for such lesions, their intrauterine growth cannot be arrested during pregnancy, and thus surgical management becomes inevitable. The lesion was excised surgically under general anesthesia using electrocautery to control bleeding. Considering the physiological immaturity of the neonatal hemostatic system, appropriate preoperative assessment, vitamin K prophylaxis, and meticulous intraoperative hemostasis are essential for safe surgical management.

Categories: Pediatrics, Dentistry
Keywords: congenital epulis, granular cell myoblastoma, granular cell tumour, histopathology, immunoreactivity

Introduction

Congenital granular cell epulis (CGCE) is a rare benign oral lesion occurring almost exclusively in newborns. It is considered a developmental lesion arising from cranial ectomesenchyme rather than a true neoplasm [1,2]. The lesion has a distinct predilection for the maxillary alveolar ridge, occurring approximately three times more frequently in the maxilla than in the mandible, with a marked female predominance [3-6].

CGCE may range from a small asymptomatic nodule to a large gingival mass that can interfere with feeding, mouth closure, and occasionally respiration. Although maternal hormonal influence has been proposed because of the female predominance, the role of estrogen and progesterone remains unproven [6-8]. Prenatal diagnosis is possible using ultrasonography and, in selected cases, three-dimensional ultrasonography or magnetic resonance imaging [9-11]. Surgical excision is generally indicated for large lesions causing functional impairment.

The differential diagnosis includes hemangioma, teratoma, neuroectodermal tumor of infancy, odontogenic lesions, and other granular cell lesions. Histopathological examination is essential for diagnosis, with CGCE typically showing large polygonal cells with abundant eosinophilic granular cytoplasm beneath stratified squamous epithelium [1].

The present case is notable for the unusually large size and atypical location of the lesion, with associated difficulty in feeding and mouth closure. This case report aimed to describe the clinical, histopathological, and immunohistochemical features of a giant CGCE in a neonate and highlight the considerations involved in its surgical management.

Case Presentation

A 10-day-old female neonate presented to our department with a large swelling arising from the anterior maxillary alveolar ridge. The lesion was associated with difficulty in approximating the lips, resulting in an almost completely obliterated oral fissure. Occasional bleeding was noted due to friction between the lesion and surrounding oral tissues. On examination, a large, soft, pink, sessile, oval, smooth, and non-ulcerated lesion measuring approximately 5 × 4 cm was noted. The lesion appeared to arise from the anterior maxillary alveolar ridge and extended into the oral cavity, mimicking a lingual outgrowth. It caused mild difficulty in breathing and swallowing, and breastfeeding was not possible; therefore, an oral feeding tube was inserted. Based on the clinical appearance, a provisional diagnosis of congenital granular cell epulis (CGCE) was made (Figure *1*).

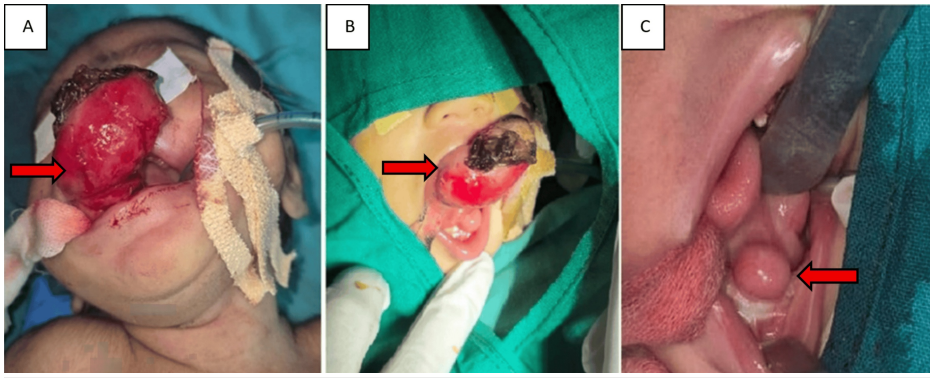


FIGURE 1: Clinical presentation of congenital granular cell epulis

(A) Large soft-tissue lesion arising from the anterior maxillary alveolar ridge. (B) Soft-tissue lesion involving the maxillary and mandibular alveolar ridge. (C) Epuloid growth involving the mandibular alveolar ridge. Arrows indicate the respective lesions.

An interdisciplinary team comprising a pediatric dentist, neonatologist, pediatric surgeon, and pathologist evaluated the patient, and surgical excision under general anesthesia was planned. Following gentle mouth opening, the intraoral component of the lesion was visualized. The mucosal surface was reddish and non-ulcerated. The lesion was excised as a single mass using electrocautery at 2-3 W to achieve adequate hemostasis. A small associated epuloid growth involving the mandible was also excised using electrocautery. The excised specimens were submitted for histopathological examination. The patient was followed up at three and 12 months after surgery, with no evidence of recurrence and normal eruption of the incisor teeth (Figures *2* and 3).



FIGURE 2: Postoperative clinical photographs following electrocautery excision

(A) Maxillary surgical site after excision of the lesion. (B) Mandibular surgical site after excision of the associated epuloid growth. Arrows indicate the respective surgical sites.

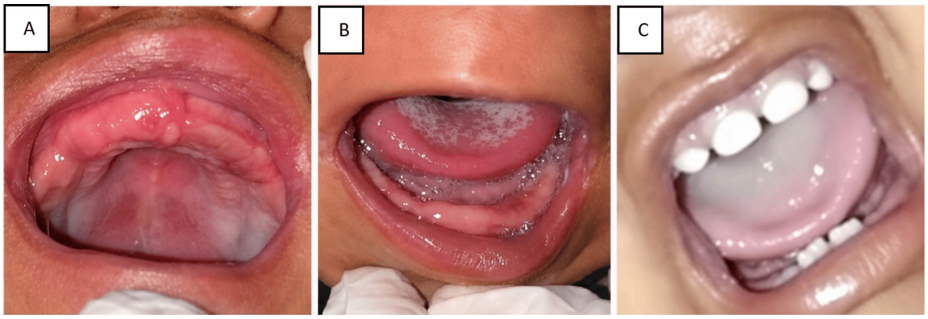


FIGURE 3: Follow-up clinical photographs showing healing after surgical excision

(A) Healed maxillary surgical site at three months. (B) Healed mandibular surgical site at three months. (C) Normal eruption of the teeth at 12 months.

Histopathological examination revealed an atrophic, non-keratinized stratified squamous epithelial lining overlying sheets and lobules of large round-to-polygonal cells with abundant eosinophilic granular cytoplasm and small central nuclei with distinct nucleoli. These findings were consistent with congenital granular cell epulis. Immunohistochemically, the lesional cells showed positivity for neuron-specific enolase and vimentin, while they were negative for S-100, laminin, chromogranin, and other tested markers, supporting the diagnosis (Figure *4*).

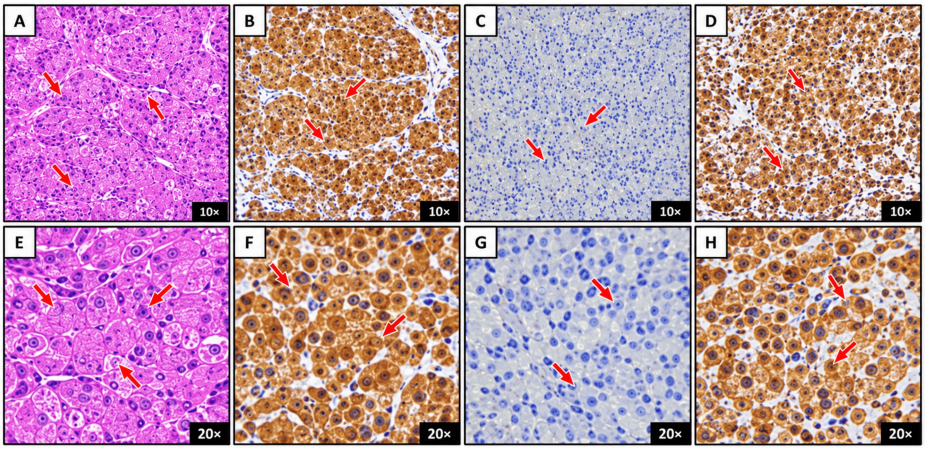


FIGURE 4: Histopathological and immunohistochemical features of the excised lesion

(A, E) Haematoxylin and eosin staining showing sheets and nests of large round-to-polygonal granular cells with abundant eosinophilic granular cytoplasm and central nuclei with distinct nucleoli at 10× and 20× magnification. (B, F) Neuron-specific enolase showing diffuse cytoplasmic positivity in lesional cells at 10× and 20× magnification. (C, G) S-100 immunostaining showing negative expression in lesional cells at 10× and 20× magnification. (D, H) Vimentin showing positive nuclear and cytoplasmic immunoreactivity in lesional cells at 10× and 20× magnification. Arrows indicate representative lesional cells.

Discussion

The major clinical concern in the present case was the neonate's ability to feed because of the unusually large size of the lesion. Despite the extensive intraoral mass, severe airway compromise was not observed. In neonates and young infants, the anatomical relationship between the epiglottis and soft palate permits simultaneous breathing and swallowing, thereby maintaining airway continuity during feeding [7]. In the present case, however, the size and location of the lesion prevented effective breastfeeding and necessitated tube feeding.

CGCE typically presents as a gingival mass arising from the maxillary alveolar ridge, usually lateral to the midline and near the primary canine or lateral incisor region [8]. The present case was unusual because the principal lesion was located centrally in the anterior maxilla, with an additional small epuloid lesion involving the mandibular gingival pad. The unusually large size and atypical distribution therefore

represented distinctive features of this case.

The pathogenesis of CGCE remains uncertain. Its marked female predominance and occurrence during fetal life have led to the hypothesis of hormonal influence; however, the absence of consistent estrogen and progesterone receptor expression does not provide conclusive evidence for a hormonal mechanism [9]. The lesion may develop during the later stages of gestation and can occasionally be detected prenatally. Ultrasonography may demonstrate a well-defined protruding oral mass as early as the second half of pregnancy, while three-dimensional ultrasonography and magnetic resonance imaging can further delineate large lesions [9-11]. CGCE may enlarge during the third trimester, with growth generally ceasing after birth.

The exact histogenetic origin of CGCE remains controversial. Current evidence supports its derivation from primitive mesenchymal cells of the cranial ectomesenchyme, which is closely related to neural crest-derived tissues involved in development of the oral and maxillofacial region [10]. Abnormal proliferation or differentiation of these cells during development of the alveolar region may contribute to lesion formation.

The differential diagnosis of a large congenital oral mass includes hemangioma, teratoma, neuroectodermal tumor of infancy, odontogenic lesions, and granular cell tumor. In the present case, histopathological examination demonstrated the characteristic sheets and lobules of large polygonal cells with abundant eosinophilic granular cytoplasm, supporting the diagnosis of CGCE. The absence of prominent vascular channels and multiple tissue components argued against hemangioma and teratoma, respectively. The absence of characteristic melanin pigmentation helped distinguish the lesion from neuroectodermal tumor of infancy. Similarly, the lack of odontogenic epithelium or ectopic tooth structures excluded odontogenic lesions [1,12]. The immunohistochemical profile, including neuron-specific enolase and vimentin positivity with S-100 negativity, further supported the diagnosis.

Another important consideration in the management of a large congenital oral lesion is perioperative hemostasis. Neonates have a physiologically immature hemostatic system, with several coagulation factors present at lower concentrations than in adults [13]. Therefore, careful preoperative assessment, appropriate vitamin K prophylaxis, meticulous surgical technique, and effective intraoperative hemostasis are particularly important when operating on neonates. In the present case, vitamin K had been administered after birth, and electrocautery was used to facilitate hemostasis during excision.

Management of CGCE depends primarily on the size of the lesion and its effect on feeding and respiration. Small, asymptomatic lesions may be managed conservatively because spontaneous regression has been reported [14,15]. However, surgical excision is preferred when the lesion causes significant functional impairment, as in the present case. Various techniques, including conventional excision and laser-assisted removal, have been described [14]. Electrocautery has also been successfully used for excision of congenital epulis while providing simultaneous hemostasis [16]. In the present case, electrocautery enabled complete removal of both lesions with satisfactory hemostasis. Follow-up at three and 12 months demonstrated no recurrence and normal eruption of the incisors, supporting a favorable outcome following conservative surgical excision.

A major strength of this case is the unusual size and atypical location of the lesion, along with comprehensive clinical, histopathological, and immunohistochemical evaluation and follow-up demonstrating no recurrence. The multidisciplinary management and use of electrocautery also provide useful practical insights into the surgical management of large congenital oral lesions in neonates. However, the main limitation is that this is a single case report, and therefore the findings cannot be generalized to all cases of congenital granular cell epulis. In addition, the long-term effect on subsequent dentition and growth could not be assessed beyond the available follow-up period.

Conclusions

Congenital granular cell epulis is a rare benign lesion that may attain a considerable size and interfere with feeding and oral function in neonates. Early recognition, accurate histopathological diagnosis, and individualized surgical management are essential, particularly in large lesions. In the present case, electrocautery enabled effective excision with satisfactory hemostasis and no recurrence during follow-up. Careful perioperative assessment of neonatal hemostasis, meticulous surgical technique, and appropriate postoperative monitoring are important for achieving safe and favorable outcomes.

Additional Information

Author Contributions

All authors have reviewed the final version to be published and agreed to be accountable for all aspects of the work.

Concept and design: Anusha Karre, Shaik Hasanuddin, K Suhasini, B Simren, Prasanna Miyapuram

Acquisition, analysis, or interpretation of data: Anusha Karre, Shaik Hasanuddin, K Suhasini, B Simren,

Prasanna Miyapuram

Drafting of the manuscript: Anusha Karre, Shaik Hasanuddin, K Suhasini, B Simren, Prasanna Miyapuram

Critical review of the manuscript for important intellectual content: Anusha Karre, Shaik Hasanuddin, K Suhasini, B Simren, Prasanna Miyapuram

Supervision: Anusha Karre, Shaik Hasanuddin, K Suhasini, B Simren, Prasanna Miyapuram

Disclosures

Human subjects: Informed consent for treatment and open access publication was obtained or waived by all participants in this study. **Conflicts of interest:** In compliance with the ICMJE uniform disclosure form, all authors declare the following: **Payment/services info:** All authors have declared that no financial support was received from any organization for the submitted work. **Financial relationships:** All authors have declared that they have no financial relationships at present or within the previous three years with any organizations that might have an interest in the submitted work. **Other relationships:** All authors have declared that there are no other relationships or activities that could appear to have influenced the submitted work.

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