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Management of congenital epulis causing feeding difficulties in an in vitro fertilized neonate: A case report

Abhishek Kumar, Sneha Shrestha, Vinay Marla

ABSTRACT

Introduction: Congenital epulis or congenital granular cell tumor is a rare benign tumor of the oral cavity presenting at birth in neonates. A slightly increased risk for congenital malformations after in vitro fertilization (IVF) persists. This condition may interfere with respiration, feeding or adequate closure of the mouth. A decisive diagnosis is made by histopathological analysis. The exact histogenesis of this lesion is still uncertain. The recommended treatment for tumor is surgical resection with no recurrences or damage to the developing dentition being reported. However, some cases have reported spontaneous regression of this tumor. Case Report: This is a case report of a 15-day-old pre-term female neonate who was in vitro fertilized and presented with a single exophytic lesion in the maxillary alveolar ridge with bleeding during feeding. The lesion was excised under local anaesthesia and confirmed by histopathological analysis as congenital epulis. The postoperative course was uneventful. Follow-up for seven months did not show recurrence. Conclusion: This is a rare case of congenital epulis in an IVF neonate. Recognition of this entity and prompt treatment is essential for preventing any difficulties for the neonate.

Keywords: Granular cell tumor, Congenital, Infant, In vitro fertilization, Newborn complications

Abhishek Kumar[1], Sneha Shrestha[2], Vinay Marla[3]

Affiliations: [1]MDS, Assistant Professor, Department of Pedodontics and Preventive Dentistry, B.P. Koirala Institute of Health Sciences, Dharan, Nepal; [2]MDS, Assistant Professor, Department of Pedodontics and Preventive Dentistry, B.P. Koirala Institute of Health Sciences, Dharan, Nepal; [3]MDS, Assistant Professor, Department of Oral Pathology, B.P. Koirala Institute of Health Sciences, Dharan, Nepal.

Corresponding Author: Dr. Vinay Marla, Department of Oral Pathology, Dhulikhel Hospital – Kathmandu University Hospital, Dhulikhel, Kavrepalanchowk, Nepal, 45200; Email: vinaymarla85@gmail.com

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INTRODUCTION

Congenital epulis, also called congenital granular cell tumor of the newborn is a rare benign gingival cell tumor that presents at birth. It was first described by Neumann in 1871 [1–3]. Two hundred and sixteen cases have been reported since then [1]. This tumor arises from the mucosa of the gingiva, most commonly from the anterior part of the maxillary alveolar ridge, and is typically seen as an exophytic mass protruding out of the newborn child's mouth, which may interfere with respiration or feeding. Congenital epulis is seen only in the newborn and is a different entity from other granular cell tumors. The risk for congenital malformations is increased in infants born after in vitro fertilization (IVF). Some specific malformations appear to be more affected than others [2].

Multiple lesions are rare, occurring in only 10% of all case reports. The size of the lesion varies from a few

millimeters to various cm in diameter with a maxillary to mandible ratio of 3:1 and a female to male ratio 8:1 [4]. The recommended treatment is surgical excision under local or general anesthesia, although spontaneous regression has been reported. There are no reports of recurrences, even if incomplete margins are excised; malignant transformation, or future disruption to teeth or gingiva [5]. There are usually no associated dental abnormalities or congenital malformations, except for occasional reports of a hypoplastic or absent tooth and the possibility of mild midface hypoplasia [6, 7]. The etiology of the condition is unknown.

CASE REPORT

A 15-day-old female neonate, born at 35th week of gestation and weighing 3.5 kg was referred to the Department of Pedodontics and Preventive Dentistry, CODS [BPKIHS] with an exophytic lesion in the anterior maxillary alveolus which was noticed at birth. The pre-term baby was conceived by test tube fertilization and born by cesarean section. Clinical examination revealed a round, firm, pedunculated mass on upper alveolar ridge, smooth surfaced with reddish color, non-tender to palpation and measuring approximately 1×1 cm in size (Figure 1A). The tumor was completely excised under local anesthesia. No other lesions or systemic involvement were found. No post-surgical complications were observed (Figure 1B).

The excised specimen was nodular, greyish brown tissue measuring approximately 0.8x0.8x0.3 cm in dimensions (Figure 1C) which on microscopic examination revealed orthokeratinized stratified squamous epithelium (Figure 2A) with the underlying connective tissue stroma showing proliferation of polygonal cells with abundant granular eosinophilic cytoplasm and round central nuclei (Figure 2B). These findings were suggestive of the diagnosis of congenital granular cell epulis. The one week follow-up of the patient was uneventful (Figure 1D). There was no sign of any recurrence after seven months follow-up.

DISCUSSION

Congenital epulis is a rare benign jaw tumor of neonate [1]. The Greek word ‘epulis’ which means ‘swelling of the gingiva’ also known as ‘congenital gingival granular cell tumor’/ Neumann’s tumor [1,3]. The lesion usually presents at birth with an obvious mass arising from the gingival mucosa, although prenatal diagnosis with ultrasound has been reported as early as 26 weeks gestation [8]. Assisted reproductive technology contributes a significant risk of congenital malformation and may be more pronounced for multiples [5].

Congenital epulis usually arises from the gingival mucosa of the maxilla or mandible (maxillary/mandibular ratio 3:1) but its occurrence has also been described on the tongue [5]. There is a marked female preponderance of 8:1 [6, 7]. The lesion is often pedunculated, flesh-pink colored, firm with a smooth or lobulated surface and in general solitary. The lesion does not increase in size after birth without any surface changes and is usually non-tender. Multiple lesions may also occur in up to 10% [4]. Size of the lesion varies from a few millimeters up to various centimeters in diameter. Large lesions may interfere with respiration, feeding or adequate closure of the mouth. Multiple tumors arising from the anterior maxillary alveolar ridge has also been reported [6]. Etiology, histopathogenesis and natural history is still unclear [8]. Diagnosis is usually made on clinical grounds alone. The treatment of choice is surgical excision and spontaneous regression of this tumor has been reported [9].

Recurrences or malignant transformation of this tumor have never been reported. No recurrences were reported in case of incomplete surgical removal. Also, damage to the underlying developing dentition due to this tumor has not been reported in the current literature [10]. There are usually no associated dental abnormalities or congenital malformations, [9] except for occasional reports of a hypoplastic or absent tooth

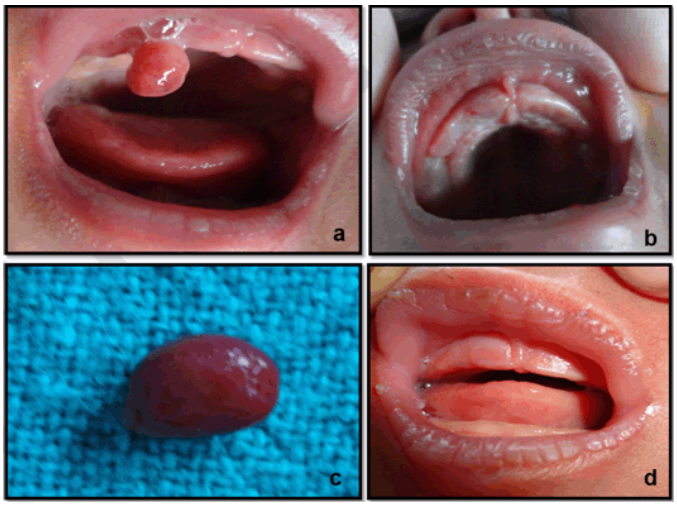

Figure 1: (A) Preoperative picture of the patient, (B) Postoperative picture of the patient, (C) Gross findings of the excised specimen and (D) One-week follow-up of the patient.

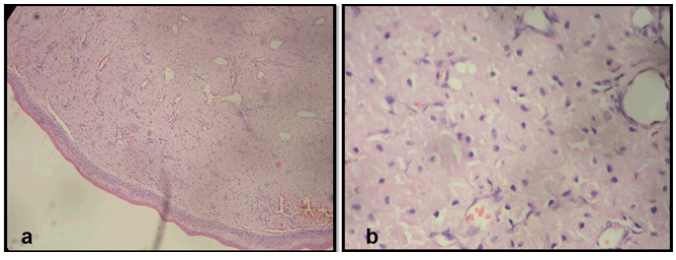

Figure 2: (A) Epithelium and underlying cellular connective tissue stroma (H&E stain, x10), and (B) Polygonal cells showing granular eosinophilic cytoplasm (H&E stain, x40).

and the possibility of mild midface hypoplasia. The etiology of the condition is unknown. Several theories have been suggested, namely, myoblastic, odontogenic, neurogenic, fibroblastic, histiocytic and endocrinologic [11–13].

CONCLUSION

Knowledge of this rare entity is essential so that such pathologies if present in a neonate can be recognized and prevent any complications and facilitate adequate feeding.

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Author Contributions

Abhishek Kumar – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published

Sneha Shrestha – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published

Vinay Marla – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published

Guarantor

The corresponding author is the guarantor of submission.

Conflict of Interest

Authors declare no conflict of interest.

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