

CASE REPORT**Neumann's Tumor: A Case Report****Kashi Nataraj Vinay¹, Lidetu Afework Anjulo^{2*}, Prabhakar Nitin³, Kashi Vinay Neha³, Dwivedi Dhara³****ABSTRACT****OPEN ACCESS**

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BACKGROUND: *The congenital granular cell tumour of the newborn, also known as congenital epulis or Neumann's tumor, is rare. It occurs on the gingiva of the anterior alveolar ridge of the jaws. This lesion behaves in a benign manner and no recurrent or metastatic lesions have been reported.*

CASE REPORT: *We are reporting a 2-day-old female neonate, who came to our unit with a well defined, solitary, firm mass arising from the maxillary anterior region measuring about 3.5 cms in diameter and causing difficulty in breast feeding but no hindrance to the airway. The mass was surgically excised under general anesthesia. Postoperative wound healing was uneventful.*

CONCLUSION: *We have shared our experience in handling this rare type of tumor. We have presented the clinical features and the different modalities of its treatment to spread awareness among clinicians for better management of similar tumors.*

KEY WORDS: *Neumann's tumor, Congenital Epulis, Congenital Granular Cell Tumor (cgct)*

INTRODUCTION

Congenital granular cell tumor (CGCT), first described by Neumann in 1871, is a benign soft tissue lesion of the neonate that almost always arises from the alveolar mucosa. It is also known as congenital epulis or Neumann's tumor (1). They are seen 3 times more frequently in the maxillary alveolus than in the mandibular alveolus (2, 3). Females are affected from 8 to 10 times more frequently than males, perhaps indicating a hormonal component in its development (4,5). The typical location is the alveolar ridge of the maxilla near the canine, but the mandibular region can also be involved. Usually, it is presented as a single lesion; however, multiple lesions have been reported in some cases (6,7). This lesion is seen as a mass protruding from the mouth of a newborn and it may interfere with feeding and respiration.

The recommended treatment plan involves prompt surgical resection of the mass under GA or LA, since spontaneous regression is rare. There are no reports showing CGCT recurrence or malignant transformation (2,5). Electrocautery and use of carbon dioxide laser

(8) have also been advocated. Some authors have also advocated gingivoperiosteoplasty with excision for Primary alveolar reconstruction in cases of defects in the alveolar ridge. It helps in achieving proper alignment and promotes normal tooth development (9). This case report intends to document the clinical presentation and its management of the Neumann's tumor in the maxillary arch of a 2-day-old female neonate.

CASE REPORT

An otherwise healthy 2-day-old neonate was referred to our Oral and Maxillofacial Surgery Unit from the Neonatal ICU of our hospital for diagnosis and treatment of a large mass protruding from her mouth. The mother was fit and well gravida 2 para 1, with no remarkable medical history to note. The infant was born by normal vaginal delivery. The baby was born at term plus eight days weighing 2.85 kg, pink and breathing spontaneously (Apgar: 9-10). As the mother was from a remote rural area, she did not have access to any antenatal ultrasonogram. At birth, a firm pedunculated mass was observed protruding from the oral cavity.

On examination, there was a solitary, firm, pedunculated mass, measuring about 3.5cms in diameter. It was arising from the upper alveolar ridge over the right lateral and central incisor area (Figure 1). There was no difficulty in respiration, but the mass interfered while breast feeding. Therefore, a nasogastric tube was passed due to concerns over feeding.

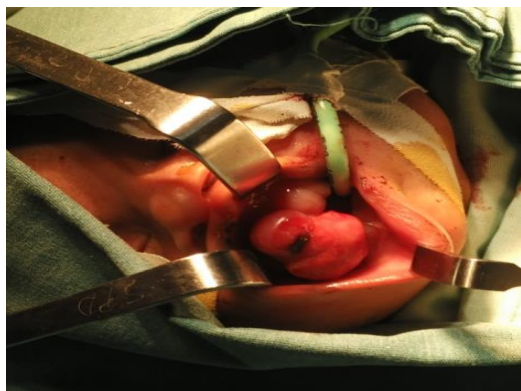


Figure 1: Pre-op view of 5th day neonate's congenital granular cell tumor

She was scheduled for surgery on the 5th day after birth. The excision of this mass was done under general anaesthesia with oral endotracheal intubation. Monopolar electrocautery was used, and there was very minimal blood loss (Figure 2). In postexcision, any kind of alveolar defect was not noted (Figure 3). Postoperative recovery was uneventful. Nasogastric tube feeding was initiated 3 hours after surgery. The child was breastfeeding 48 hours after surgery, and she was discharged the following day. Healing was uneventful, and the gingival reepithelised in 10days. Histopathology revealed focal ulceration with underlying stroma demonstrating large sheets of closely packed, polygonal cells with round, regular nuclei and inconspicuous nucleoli. It also showed abundant granular cytoplasm consistent with the diagnosis of congenital epulis, or congenital granular cell tumor (Figure 4).



Figure 2: Excised congenital granular cell tumor

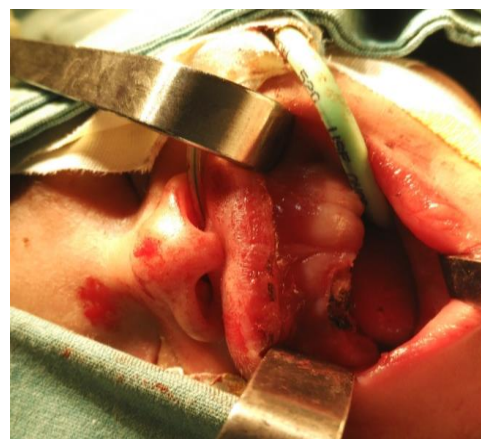


Figure 3: Post-op view of the 5th day neonate after excision congenital granular cell tumor with cautery

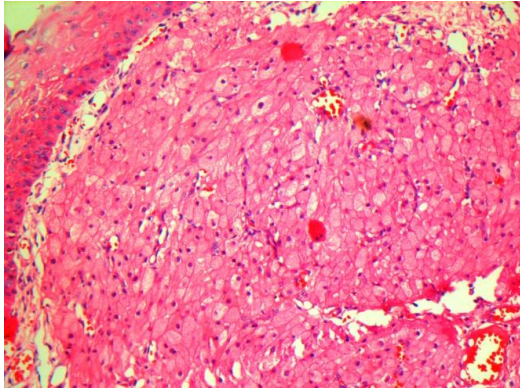


Figure 4: Histopathology of excised congenital granular cell tumor

DISCUSSION

Congenital epulis, also known as granular cell tumour of the gingiva, congenital granular cell myoblastoma, or Neumann's tumour, following the first published case (1), is encountered exclusively in newborns. The etiology remains unknown and controversial. Congenital epulis differs from other granular cell tumours encountered in adults by its exclusive origin from the neonatal gingiva, the scattered presence of odontogenic epithelium, the more elaborate vasculature and the lack of interstitial cells with angulate bodies (10, 11) different entity from other GCTs. The tumor has a marked female preponderance of 8:1. The tumor usually arises at the future site of the maxillary canine or the lateral incisors, but the unerupted teeth are not involved. It has been described as arising from the mandibular gingiva as well as from several other locations simultaneously. The reported size varies from several millimeters to 7.5 cm⁽⁸⁾.

The clinical presentation consists of a lobular or ovoid, sessile or pedunculated swelling covered by a smooth mucosal surface, usually in the maxilla. A provisional diagnosis is often made clinically at birth and is confirmed histologically. The differential diagnosis of a large mass in the fetal or neonatal oral cavity should include such congenital malformations as encephalocoele, dermoid cysts or teratoma and benign and malignant neoplasms including hemangioma, lymphatic malformations,

melanotic or pigmented neurectodermal tumours of infancy and rhabdomyosarcoma (10).

The treatment of choice is surgical excision to be performed as soon as possible (12). The use of general anesthesia seems to be the recommended procedure in such cases. There are reports of lesion removal without the use of anesthesia. However, this kind of procedure is not justified since these surgical interventions are elective. The CGCT removal under local anesthesia is also an alternative (7,8,11) when intubation is not possible or in cases of small lesions. Some wait for spontaneous regression of small lesions. There is also the possibility of removal during the delivery in cases where the lesion was detected during pregnancy (12). This approach may eliminate additional procedures such as anesthesia and intubation, and it provides the newborn with a free airway and an unobstructed oral cavity immediately after birth (7). Recurrence after removal is not seen, infant recurrence is not seen even after incomplete excision (13). Excision of these masses can leave a notch in the alveolus that may result in an incomplete dental arch.

Gingivoperioseoplasty is performed early to correct it so that it can restore physiological continuity across the premaxilla and allow osteogenic hematoma formation between the anterior maxillary cleft. It is thought that the union of the mucoperiosteum across an alveolar and anterior hard palate defect creates a periosteal tunnel conducive to bone formation and normal tooth eruption along the cleft region (9,14).

In conclusion, Neumann's tumor or congenital epulis is a very imposing tumor of the oral cavity in neonates. It can be alarming for parents and clinicians. The tumor is often misdiagnosed before surgery because of its rarity and lack of awareness among clinicians (15).

Neumann's tumors rarely cause compromise in the airway, but most of them do hamper breast feeding. It does not harm the future dentition. It is ultimately a benign lesion and does not recur postsurgical excision. Therefore, radical resection is not warranted.

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