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Case Report

Congenital epulis of newborn - A case report

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ABSTRACT:
Congenital Epulis (CE) of the newborn is a rare benign tumor of the soft tissue which normally affects the maxillary alveolar ridge in the neonates. The treatment of choice is surgical excision, due to its possible interference with feeding, respiration or inadequate closure of the mouth. In the present article, a case report of an otherwise healthy female neonate with CE is presented. The soft tissue tumor protruding out from the neonate's mouth was found to be attached to the right maxillary alveolar ridge in the canine region. It was successfully treated by surgical excision under general anesthesia. A 6-month follow-up of the patient showed normal healing of the mucosa and no recurrence of thelesion.
Keywords: Congenital epulis, Tumor, Newborn, Neonate, Resection.

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INTRODUCTION

Congenital epulis (CE) or congenital granular cell tumor of the newborn is a rare benign tumor of the soft tissues, which normally affects the maxillary alveolar ridge in neonates. Neumann first described CE in 1871, hence it is also known as Neumann's tumor **(Neumann E etal.1993).**

The exact nature of this entity is not clear, however, it is now thought to originate from primitive mesenchymal cells of the neural crest **(Zarbo RJ et al. 1983).** Due to its unclear etiology, pathogenesis and origin several theories have been proposed to explain its pathogenesis. These are the myoblastic, odontogenic, neurogenic, histiocytic and endocrinologic theories **(Kannan SK et al. 2006).**

The treatment of choice is surgical excision, due to its possible interference with feeding, respiration **(Chindia ML et al. 1994)** or inadequate closure of the mouth (McGuire et al. 2006). Although a few cases of spontaneous regression have been reported; however, this occurred in lesions that were very small in size **(Abdelmonieum M. et al 2005)**.

CE has a good prognosis after surgical excision as it is a benign lesion with solitaire occurrence **(Jenkins et al. 1989)** and almost no recurrence after resection **(Kaumae et al. 2015)**.

The lesion most likely develops late in utero as it is often not detected on antenatal ultrasound. Some prenatal conditions that may cause polyhydramnios, a medical condition describing an excess of amniotic fluid in the amniotic sac that might led to the formation of CE **(Eghbelian F et al. 2009), (Adeyemi BF et al 2010).**

CASE REPORT

The aim of this case report is to present a case of a 27 day-old female with intraoral mass that was noticed by the parents at birth, who reported to Punjab Government Dental College and Hospital, Amritsar, Punjab. **(Figure-1).**

A full-term baby girl with APGAR scores of 9/10 at 1 and 5 minutes and a birth weight of 3500 g was born by normal vaginal delivery to a healthy 25-year-old mother after her secondary normal pregnancy. On

physical examination, a soft tissue tumor protruding out of newborn's mouth was found attached to the right maxillary alveolar ridge in the region of canine(Figure-2).

It was well-circumscribed, smooth surfaced and red in colour much like alveolar mucosal tissue. There was no tenderness or surface change and the lesion did not increase in size after birth. The mass prevented normal closure of the mouth and interfered with breastfeeding. The mass posed no immediate airway concerns to the patient. **(Figure-2).**

The possible diagnosis was congenital epulis of newborn. Apart from that, no other symptom was reported for this neonatal patient. There was no family history of trauma, chronic irritation or any congenital abnormalities. The newborn was otherwise healthy: hematologic (complete blood count, partial platelet, C-reactive protein, erythrocyte sedimentation rate, prothrombin time, partial thromboplastin time, bleeding time) and chemical blood analysis (Na, K, BS, Ca, Crt) were within normal limits.

Patient's parents were given an oral explanation and a detailed informed consent form was signed by them according to the Helsinki declaration. The tumor which was considered to cause oral feeding problems due to obstruction was planned to be excised surgically under general anesthesia with oral intubation (Figure-3,4).

The lesion was gently pushed to the side, the airway was visualized and an oral endotracheal tube was inserted. The lesion was completely excised under general anesthesia followed by suturing **(Figure-5)**. The resected mass measured 1cm in size **(Figure- 6).** Postoperative instructions were given to the patient's mother. The intraoperative and postoperative course was uneventful. The newborn recovered with no complications. Regular oral feeding was initiated immediately after surgery and was well tolerated.

After 1 week, the sutures were removed and normal post-surgical healing was observed **(Figure-7).** The excised mass was sent for histologic examination and it confirmed the pre-diagnosis of congenital epulis showing a proliferation of round cells with a finely granular eosinophilic cytoplasm with round, fine nucleolus in the nuclei, with no signs of atypia or mitotic activity **(Figure-8).** A 3-month and 6-month follow-up showed normal healed mucosa and no signs of recurrence of the lesion **(Figure-9, 10).**

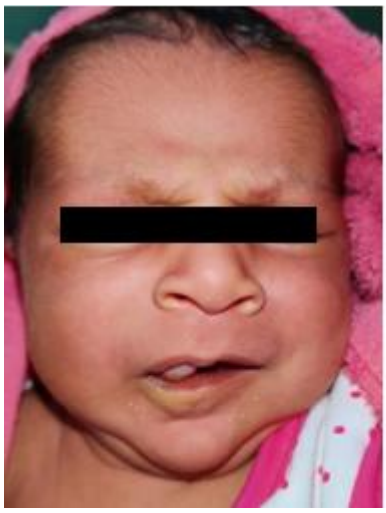
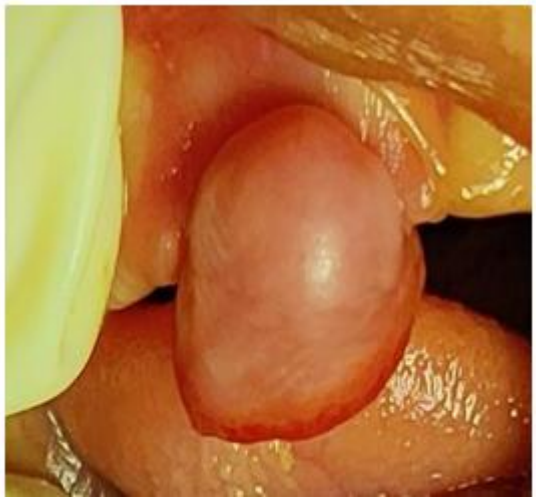


FIGURE 1



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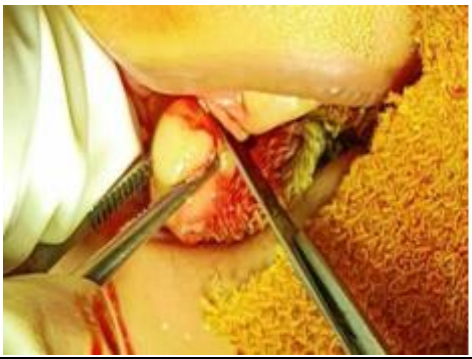

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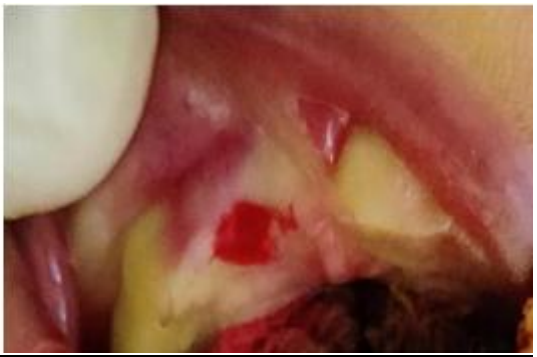

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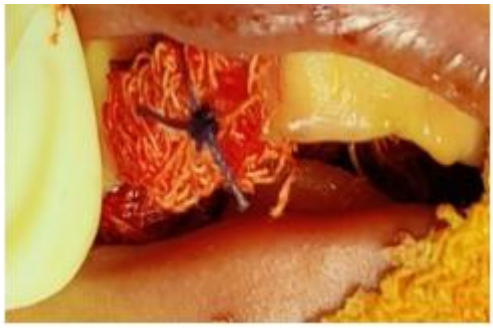

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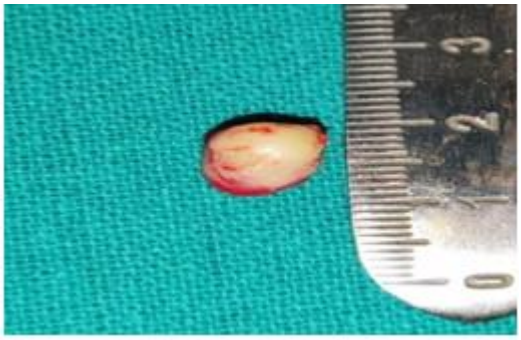

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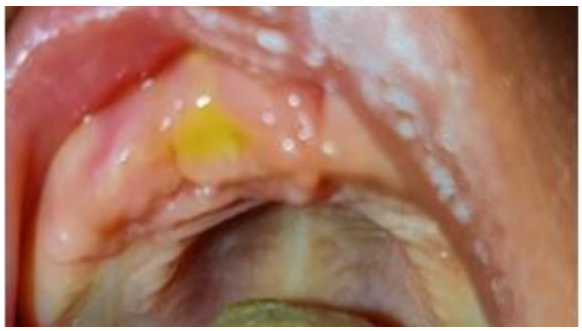

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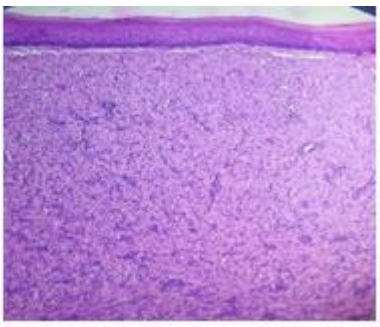

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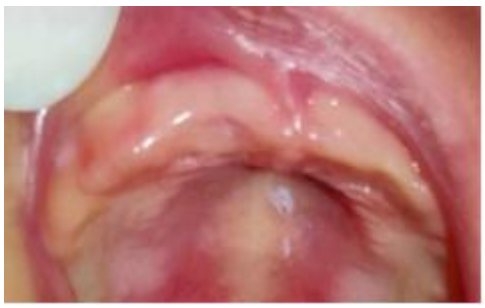

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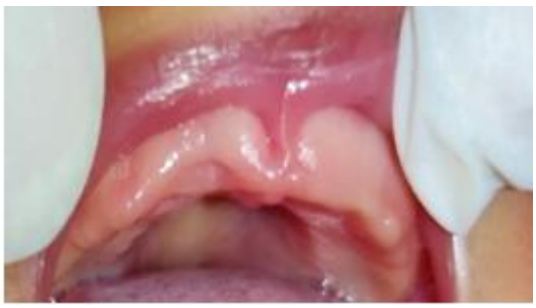

FIGURE10

DISCUSSION

CE is also known as congenital myoblastoma, gingival granular cell tumor or Neumann tumor **(Ben H. et al. 2010).** CE has been reported with an 8:1 to 10:1 female: male ratio as documented by **Inan M et al. (2002).** The cause of female predisposition might be due to the possibility of an intrauterine stimulus from the fetal ovaries as hypothesized by **Subramaniam R. et al (1993).** Likewise, our case was also a 27-day old female neonate, with the same condition.

This condition clinically appears as a protuberant round or ovoid mass, pedunculated or sessile, which may interfere with respiration and feeding, as seen in our case. CE usually occurs in the mucosa of the maxillary alveolus as smooth surfaced pink mass, frequently lateral to the mid line in the area of the developing primary lateral incisor and canine as documented by **Bilen et al. (2004) and Inan et al. (2002).** Similar findings were present in our casetoo.

It is usually not associated with any other abnormality of the teeth or other congenital abnormalities. This tumor is a rare anomaly with only 0.0006% incidence as documented by **Bosanquet D. et al. (2009).**

It has similar appearance to Granular Cell Tumor, but there are several distinguishing features of GCT, such as predilection for newborn females, anterior maxillary location **(Chami RG et al. 1886)**, presence at birth, plexiform arrangement of capillaries, and lack of pseudoepitheliomatous hyperplasia **(Bork M. et al.1996)**.

It usually occurs as a single mass although 10% cases occur as multiple lesions with the size of the lesion varying from few millimeters to 9 cm. Similarly, in our case, a single mass was present which was 1 cm in size and it was affecting oral feeding of the 27-days old neonate.

It is important to stress that clinicians should know the differential diagnosis of different growths in the oral cavity of newborns, including hemangioma, lymphangioma, fibroma, granuloma, rhabdomyosarcoma and osteogenic and chondrogenic sarcomas, as treatment modalities will be different for each case.

It has similar appearance to Granular Cell Tumor, but there are several distinguishing features of GCT, such

as predilection for newborn females, anterior maxillary location **(Chami RG et al. 1886),** presence at birth, plexiform arrangement of capillaries, and lack of pseudoepitheliomatous hyperplasia **(Bork M. et al. 1996).**

In the present case, the removal of CE can be done by surgery under local or general anesthesia, electrocautery, and carbon dioxide laser set at 15W continuous wave. In our case, the tumor was resected by surgical excision under general anesthesia as done earlier by **McGuire TP. et al. (2006).**

Recurrence of the lesion, damage to the adjacent tissues or any malignant transformation has not been reported in literature so far **(Merrett SJ et al. 2003), (Dhingra M et al. 2010).**

A 6-month post-surgery follow-up of our case has shown normally healed mucosa and no recurrence of the lesion.

CONCLUSION

In conclusion, CE is a neonatal congenital tumor with very rare occurence. The treatment of CE is surgical excision. If early treatment is not executed, tumor may cause difficulties in oral feeding and respiration. Recurrence of the tumor and damage to future dentition have not been reported.

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