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Congenital Epulis of the Newborn: A Case Report

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Abstract

Background: Congenital epulis (CE) also known as congenital granuler cell tumor is a rarely encountered pathology the majority of which originates from the gingival mucosa, particularly the anterior portion of the maxillary alveolar ridge. CE mostly seen in girls. CE with unclear histogenesis and etiology is seen at birth as a solitary mass in oral cavity. Apart from non-congenital epulis, it contains granular cells. So lesion is named congenital granular cell tumor. CE has a benign histopathology and after surgery there is no recurrence reported in the literature. **Aim:** The purpose of this case report, is to present, 5 day-old female neonatal girl who was seen CE on the left maxillary alveolar ridge on the region of the future incisors. **Case Presentation:** The tumoral lesion was well-circumscribed and 10 mm in diameter, smooth surfaced and red in colour much like alveolar mucosal tissue. Tumoral lesion was affecting oral feeding due to obstruction. Excisional biopsy was performed under topical anesthesia. The histopathology was reported as congenital epulis. During the 4 months follow-up, we have seen no complication. **Conclusion:** CE is a neonatal congenital tumor which is very rare. The treatment of CE is surgical excision. Unless the early treatment is not executed, tumor may cause difficulties in oral feeding and respiration. Therefore it should be excised in an early period.

Keywords

Newborn, Congenital Epulis, Neonatal

1. Introduction

Congenital epulis (CE) is also known as congenital granular cell tumor; congenital gingival Neumann's tumor and congenital myoblastoma was firstly

defined by Neumann in 1871 [1] [2] [3]. This very rarely encountered soft tissue tumor in newborn is generally located in maxilla [4]. The diagnosis is often made postnatally during neonatal care. However, prenatal diagnosis is important to decide on the route of delivery and plan early multidisciplinary postnatal management [5]. They are mostly pedunculated lesions and particularly located in the area of the incisors. CE is seen in girls at the ratio of 80% and its histopathology table is like granular cell myoblastoma but pseudoepitheliomatous hyperplasia is not seen in it.Large cells that have eosinophilic cytoplasm forms parenchyma of tumor [6]. CE has a benign prognosis and almost never recurrences [7]. Most of CE are soliter nevertheless there are cases that are large and multiple. The treatment of choice is surgical excision, although a few cases of spontaneous regression have been reported; however, this occurred in lesions that were very small in size [8] [9]. Aim of the this case report is to present a 5 day-old female with CE on the anterior maxillary alveolar ridge who is treated in our clinic.

2. Case Report

A 5-day-old female neonate presented to our hospital on account of a congenital intraoral mass that was noticed by the parents at birth. On physical examination, a soft tissue tumour protruding out of newborn's mouth was found attached to the left maxillary alveolar ridge on the region of incisors. It was well-circumscribed and 1 cm in diameter, smooth surfaced and red in colour much like alveolar mucosal tissue.The possible diagnose was congenital epulis (**Figure 1**). Apart from that, any other symptom was not reported for this neonatal patient. As the result of the anamnesis, it was noted that the patient had no systemic disease and her mother had cesarean delivery after a normal pregnancy period. There was no family history of trauma, chronic irritation or congenital abnormalities. Patient's parent was given an oral explanation and detailed informed consent form was signed by patient's parent according to the Helsinki declaration. The tumor which was considered to cause oral feeding problems due to obstruction excised totally from its narrow pedicle by scalpel under topical anaesthesia. After excision alveolar defect was left for secondary wound healing. Hemorrhage was minimal during operation and there was no complication (**Figure 2**). The histological examination confirmed the pre-diagnosis of congenital epulis showing a proliferation of round cells with a finely granular eosinophile cytoplasm with round, fine nucleolus in the nuclei, with no signs of atypia or mitotic activity. The tumor was covered by a typical epithelium of the gingival mucosa (**Figure 3**). At the 1st and 4th month controls it was seen that operation area healed without any complications in the neonatal patient (**Figure 4** and **Figure 5**).

3. Discussion

CE is a benign tumor and very rarely encountered in newborns.This tumor is also known as congenital myoblastoma, gingival granular cell tumor or Neumann

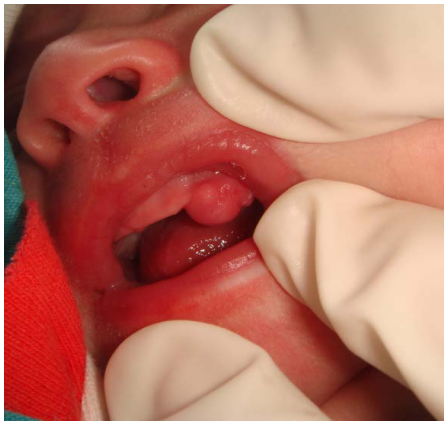

Figure 1. A tumoral lesion that is 1 cm diameter, round, pedunculated, covered with mucosa on the left maxillary region in a 5 day-old neonatal girl.

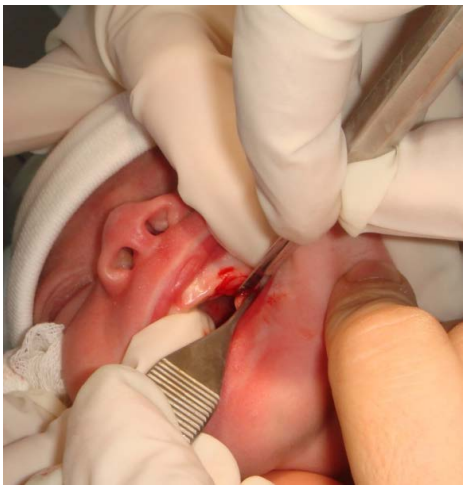

Figure 2. Minimal alveolar hemorrhage after total excision of tumoral formation.

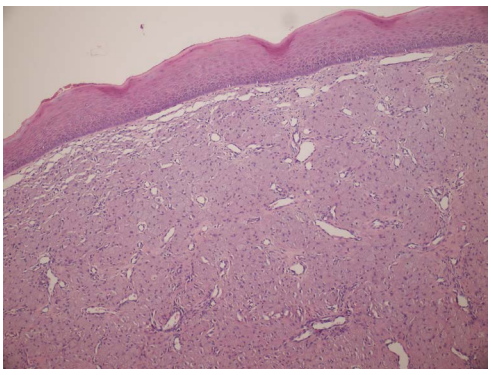

Figure 3. Histological aspect: granular cell tumor covered with typical epithelium of the gingival mucous membrane and cells with large, eosinophilic, granular cytoplasm and small nuclei (HE × 100).

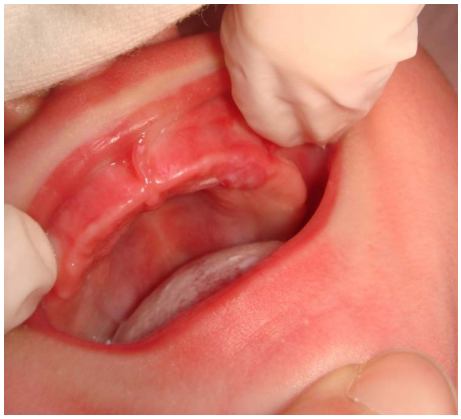

Figure 4. 1st week post-operation view.

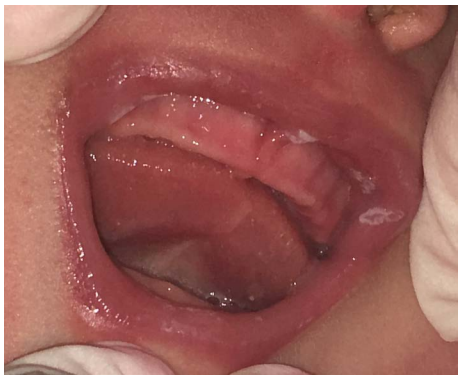

Figure 5. 4th month post-operation view.

tumor [10]. Due to its unclear etiology, histogenesis and origin several theories have been proposed to explain its histogenesis. These are the myoblastic, odontogenic, neurogenic, histiocytic and endocrinologic theories [2] [11].

CE which in general originates from the alveolar crest of the maxilla is reported three times more frequent in the maxillary alveolus than in the mandibular alveolus [12] [13]. Although the histology and the origin of CE are not known yet, the electron microscopic examination of CE shows evidence that it originates from gingival stromal cells [4]. Neonatal patient who is treated in our clinic also had CE located in maxilla.

Any recurrence or malign transformation hasn't been reported in the literature [2] [14] [15]. CE was reported to be such a benign lesion that even it still keeps being residive after excision wouldn't recurrence moreover it would spontaneously regress [14] [15]. After controls we also didn't find any evidence of recurrence in our case.

CE is commonly seen in girls. Rate of incidence is reported to be 8:1 to 10:1 female-to-male ratio. An endogenous hormonal influence has been proposed to explain this gender bias, but this has not been supported by detectable estrogen and progesterone receptors within the lesion [3] [15] [16]. Another theory to explain this female preponderance is the possibility of an intrauterine stimulus from the fetal ovaries [17]. In our case it was seen in a 5-day old neonatal girl.

CE may effect muscles that close the mouth and may prevent swallowing that makes oral feeding impossible. Especially in antenatal period if size of the lesion is large, respiration may be effected and this may cause polyhydromiosis [1] [2] [9]. The diameter of CE varies between 0.1 cm to 4 cm. The largest tumor ever reported is 7.5 cm in diameter [18]. In our case CE was 1 cm in size and despite it had no negative effect of respiration it was effecting oral feeding of the 5-day old neonatal girl.

This tumor occurs usually as a single mass, but it has been reported that 5% - 16% of cases may be as multiple. When the tumor occurs as multiple, maxilla or mandible are the most frequent locations [18] and the tumor may cause respiratory obstruction [3]. In our case CE was as a single mass and there was no respiratory distress but there was oral feeding problem.

In one case congenital goiter with CE and in another case midfacial hypoplasia with CE were reported. In our case, the patient had no other congenital anomaly, no medical history, not even the occasional midfacial hypoplasia and anomalies of the nasal complex that is sometimes seen in some cases due to pressure effect from the lesion on the developing maxilla [11].

The lesion was attached by means of a thin pedicle to the alveolar ridge. This is in consonance with most cases that are pedunculated [2] [10] [14]. Reported cases have been excised under local anesthesia as well as under general anesthesia [9] [11] [19]. In our case, the lesion was excised under topical anesthesia because of the pedicle of the mass is thin.

The diagnosis of CE should essentially be clinical in fact, prenatal imaging of congenital epulis is possible by ultrasound and MRI, but unfortunately this oral tumour has rarely been diagnosed prenatally [20] [21] [22]. In our case, patient had no prenatal follow-up.

4. Conclusion

In conclusion, CE is a neonatal congenital tumor which is very rare. The treatment of CE is surgical excision. Unless the early treatment is not executed, tumor may cause difficulties in oral feeding and respiration. Therefore it should be excised in an early period.

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