A premature newborn with intraoral tumor

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Case

A baby born by normal spontaneous vaginal route with a gestational age of 32 weeks and 4 days as the third child of Syrian parents who were first cousins was hospitalized in our neonatal intensive care unit because of respiratory distress and intraoral mass. The body weight of the female patient was found to be 1 789 g (10-50 percentile) and her head circumference was found to be 29 cm (10-50 percentile). On physical examination, the body temperature was measured to be 36.7°C (axillary), the blood pressure was found to be 55/30 mmHg, the apical heart rate was found to be 148/min, the respiratory rate was found to be 75/min and oxygen saturation was found to be 83%. The lung sounds were decreased on auscultation in the baby who had groaning, tachypnea and intercostal retractions and an oval, solid mass with a size of 2 x 3 cm localized anteriorly which nearly completly obstructed the mouth space and which was bound to the maxillary gingiva from which it originated with a fibrous handle of approximately 0.5 cm was present in the upper chin of the baby (Figure 1). Reticulogranular appearance was present on lung graphy of the patient who was ventilated mechanically because of respiratory failure and respiratory acidosis. One dose of surfactant was administered to the baby who was diagnosed with respiratory distress syndrome. After screening for sepsis was performed, ampicillin and gentamycin treatment was started with suspicious congenital pneumonia. Hemogram and C-reactive protein values at presentation were within the normal limits.

On the postnatal third day, the intraoral mass was excised from the gingiva which it was originated from with the help of a monopolar cautery under general anesthesia.

The patient who did not develop complication in the postoperative period and whose wound healing was not problematic was extubated on the next day. The patient was monitored on nasal continious positive airway pressure (CPAP) for one day and had no need for oxygen on the 10th day after delivery. Cranial and abdominal ultrasonography and echocardiography performed for screening anomaly were found to be normal. On the postnatal 18th day, the patient was completely fed orally and was discharged on the postnatal 32nd day with a body weight of 2 270 g. No recurrence occured in the three-month follow-up period.

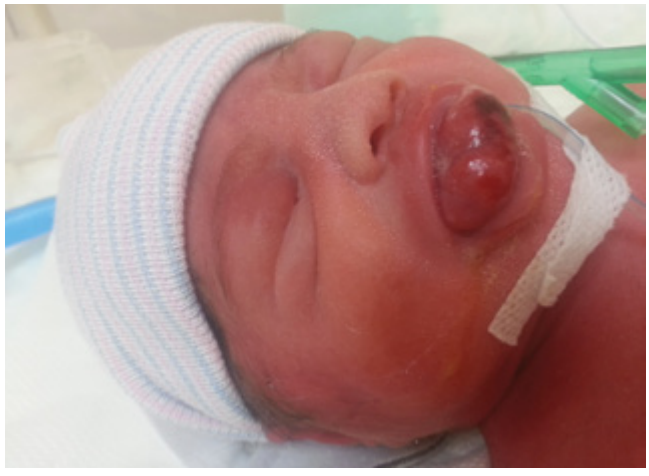

Figure 1. Mass obstructing the mouth space nearly completely

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Diagnosis: Congenital epulis

The mass which had a short handle, erythematous, punctate minimally hemorrhagic areas and a moderately firm consistency was sent for histopathological examination. On macroscopic examination, a cream-brown lesion covered with mucosa with dimensions of 2.5 x 2.1 cm was observed and hemorrhage was observed on the surface. The cross sections were solid, brown and elastic. On microscopic examination, a nodular lesion covered with stratified squamous epithelium with diffuse ulcers on the surface which was characterized with small monotonous poligonal cells with large, granular, eozinophilic cytoplasms was observed. These histopathological findings were compatible with congenital epulis (CE).

Discussion

Congenital epulis is a rare, benign soft tissue lesion of the newborn generally originating from the alveolar prominence of the maxilla or mandible. CE which is also called congenital granular cell tumor (GCT) was described by Neumann (1) in 1871 for the first time and hundreds of cases have been reported until the present time. Its characteristic property is that it generally originates from the upper chin and has a single, polypoid structure. However, multiple masses have been reported in 10% of the cases (2). Its etiology has not been elucidated yet. It is observed in girls with a higher rate compared to boys (3). The maxilla/mandible ratio is 3:1 and the female/male ratio is 8:1 or 10:1. Although its higher prevalence in girls suggested hormonal impact, the fact that estrogen and progesteron receptors could not be found on tumor cells confuted this assumption. Although malign forms of other GCTs have been observed, no malignancy related with CE has been reported in the literature so far (2, 4). Although spontaneous reduction has been reported in the literature, its treatment generally consists of complete excision of the mass. No recurrence or metastasis has been reported after surgery. The prognosis is excellent (2, 5, 6).

Premature delivery has been reported very rarely in hundreds of patients with congenital epulis in the literature (7, 8). The fact that our patient was delivered prematurely is the speciality of our case.

Congenital epulis for which the biological origin has not been elucidated yet is histologically observed as cell groups in diffuse layers. These cell groups contain round, intense small nuclei and rough granular cytoplasms. Fine vascular plexus is found between the granular cells. This predisposes to hemorrhage in the lesion (9). It is thought that congenital epulis originates from undifferentiated mesenchymal cells, fibroblasts, myofibroblasts, Schwann cells or odontogenix epithelial cells (10).

Congenital epulis causes to problems which require urgent intervention related with respiration and nutrition and severe parental anxiety because of the region of origin. Congenital epulis may render feeding impossible by preventing closure of the mouth and swallowing. This swallowing disorder leads to polyhydramniosis in the prenatal period (11). The diagnosis is frequently made after delivery, but cases diagnosed in the prenatal period have been reported in the literature (12, 13). Prenatal diagnosis is significant in terms of deciding the mode of delivery and early multi-centered approach. Its treatment is complete excision of the mass as we performed in our patient.

Congenital epulis has not been associated with any genetic disease or syndrome until the present time (2). No pathological finding was found in the screening of anomaly in our patient. However, it has been reported that large masses may lead to facial growth retardation in the middle line and anomalies in the nose and maxillary region (14). Our patient had no facial anomaly in the middle line.

Premature delivery has been reported very rarely in hundreds of congenital epulis cases in the literature. This premature baby born with congenital epulis was presented to share our experience with physicians and review the literature.

Informed Consent: Written informed consent was obtained from patient's parents via translator.

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