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CONGENITAL EPULIS (A CASE REPORT)

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Summary: Congenital epulis (CE), or granular cell tumor of the newborn is a rare, benign tumor which occurs mainly on the maxillary ridge, with predominantly in female infant. A review of the medical literature published in English revealed that the involvement of both jaws is uncommon. A female infant with simultaneous lesions on both jaws, is presented.

Key Words: Congenital epulis, granular cell tumor

The congenital epulis usually appears as a predunculated, smooth surface mass, mainly on the anterior maxillary ridge, with predominantly in female infants (5). It is a rare benign granular cell tumor of the gingiva. Since its first description by Neumann (10), more than 164 cases have been reported (15). Its size varies with range from a few milimeters to several centimeters, If large enough, it may cause nursing and respiratory problems, therefore early surgical excision is indicated(1). Only one recurrence have been reported(4), and there is no evidence for interfering with tooth eruption and development(2).

In this report, we present an unusual case of congenital epulis with simultaneous lesions on both jaws.

Case Report

A 1-day-old female infant was referred to the Plastic Surgery service, with a protruding mass from the oral cavity. The tumor was present at birth and interfering with feeding. Weighing 3400 g child was born at full term pregnancy and a normal delivery. There was no other abnormality in general examination and laboratory tests were normal. Intraoral examinatiion revealed a white colored, large mass attached to the upper left alveolar ridge, in the canine region. Medial to this tumor, there was a smaller pinky mass. Another much smaller mast was found in the left incissor area, on the mandible. They had sessile attachments to the gingiva (Fig.1). All the tumors were excised at the bases, under general anaesthesia with elliptical incision which is made up to the periosteum, and bleeding from the beds of tumors were controlled by electrocautery. The excision sites were packed with surgical spongens.

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Figure1. Congenital epulis, involving both maxilla and mandible.

On gross examination three well demarcated tumors measured 3x2 cm, 0.5X0.5 cm, and 0.4x0.4 cm respectively (Fig. 2). The cut surface of the smaller maxillary tumor was reddish-pink, and the other was white-gray in clor, microscopically the tumors were covered with normal stratified squamous epithelium without rete ridges or hyperplasia(Fig. 3). The tumors consisted of large polyhedral cells in sheets with abundant granular acidophilic cytoplasm. The nuclei were small, centrally or eccentrally placed without mitotic figures or pleomorphism. Vascular channels were numerous, especially in the smaller maxillary tumor (Fig. 4). Follow up at 6 months showed no evidences of recurrence at the excision site.

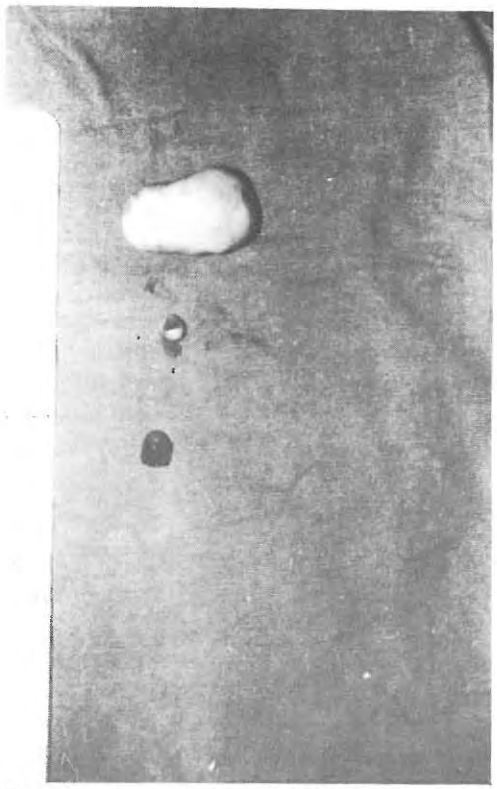

Figure 2. Three excised tumors on gross examination. The smaller mass was located on the mandible.

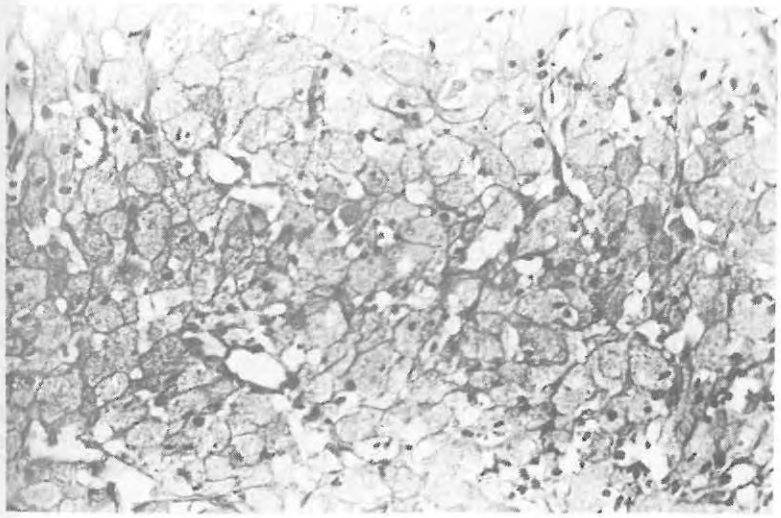

Figure 3. Typical histological appearance of the congenital epulis (HEx75).

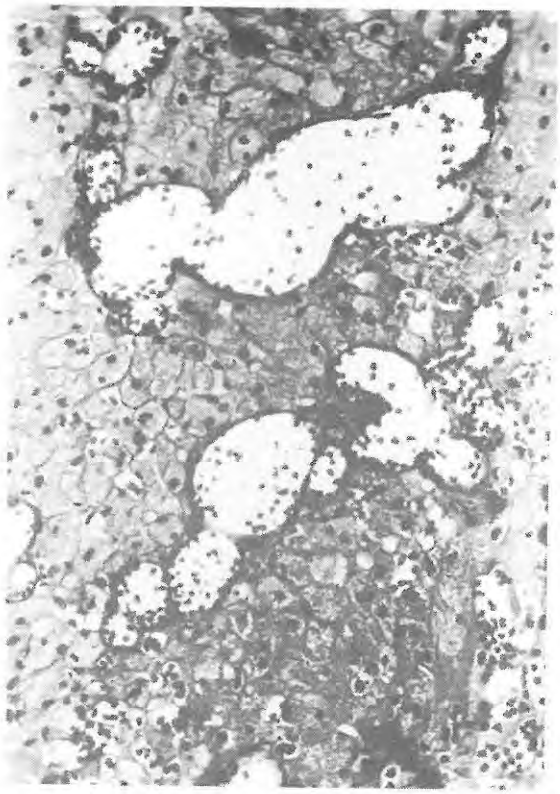

Figure 4. High power histological appearence of the more vascular smaller maxillary mass (HEx100).

Discussion

In about two thirds of cases, CE were occured on the maxiallary ridge (5). Its occurence on the both jaws seems to be extremely rare. Delaire at al (4), Kay et al (6), O'brein and Pielou (11), Blair and Edwards (1) and Uglesic and Bogati (14) reported such cases. O'brein and Pielou (11), left infact the maxillary mass without surgical intervention and it had resolved spontaneously which was smaller then mandibular mass. They concluded that spontaneous regression of the tumor may be seen in small ones. In our patient, the large tumor was interfering with infant's nursing so we removed all tumors for patients care and sent to the pathological examination.

The main histologic features of the CE are not controversial but its Histogenesis is still unclear. Fuhr and Krogh (5) reviewed the five principal theories of origin, as odontogenic, fibroblastic, histiocytic, neurogenic and myeloblastic. Ultrastructural studies have been started by Kay et al (6). They have considered an odontogenic epithelial derivation. The others proposed that CE originates from different cells while all supported the mesenchymal origin. THe undifferantiated mesenchymal cells, pericytes, stromal cells, and mesenchymal cells with potential for smooth muscle differentiation were proposed for origin of the tumor cells (7,12,13,16). There are some similarities between CE and granular cell myoblastoma but differatial diagnosis is made easily. CE occurs at birth, with female sex preponderance and it usually occurs in the maxillary ridge. It is highly vascular and its epithelium lacks pseudoepitheliomatous hiperplasia(3,16), demonstrated that the granular cells of CE, unlike of the granular cell myoblastoma, did not stain with antiserum to S-100 protein. The same result was obtained by others (8,9). They also described S-100 protein positive isolated nongranular cells which disturbuted at the periphery of some blood vessels in indeterminate nature, in their reports. Although all these studies, the histogenesis of CE is still unclear. In our case, we did not find any possibility of study for ultrastructural and immunohistochemical. The new cases making ultrastructural and immunohistochemical studies will provide more evidence to the histogenesis of this tumor.

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