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Multiple Congenital Epulides in A Newborn: With Special Reference to Reported Cases in Japan

Takenori ISHIKAWA, Taketomi NAKAI, Kinji FUJIMOTO,
Tadashi HARADA, Junji TABUCHI and Tsunehiro SHIMOSATO

Department of Oral and Maxillofacial Surgery II, Hiroshima University School of Dentistry, 1-2-3, Kasumi, Minami-ku, Hiroshima 734, Japan

ABSTRACT

This report describes a case of multiple epulides which simultaneously occurred on the maxilla and the mandible of a 6-day-old female baby. The clinical findings, diagnostic procedure, surgical treatment, histopathology, and 2-year follow-up of this case are presented. Based on the clinicostatistical and histopathologic investigations with special reference to the pertinent Japanese literature, a possible histogenetic etiology of this entity is discussed. Any odontogenic component might play an important role in the developoment of this tumor. This case of multiple epulides is considered to be very rare.

Key words: *Congenital epulis, Multiple, Histogenesis*

Congenital epulis is a rare soft tissue tumor which occurs on the alveolar ridge of the newborn. So-called congenital epulis has been generally regarded as a granular cell tumor. Since it was first described by Neumann[14], there have been scattered case reports of this tumor. The lesion occurs more often in the alveolar region of the maxillary anterior tooth than in that of the mandibular tooth. It appears as a pinkish, pedunculated round mass without ulceration. Most of the cases are solitary, but few being multiple or multilobular. A rare case of multiple congenital epulides involving both the maxilla and mandible is reported with a review of the pertinent Japanese literature. Moreover, a possible histogenesis of this entity is suggested.

CASE REPORT

Patient: The case is a 6-day-old female baby born after full-term pregnancy. Weight at birth was 2988g. No abnormalities other than tumors of the oral cavity were found on physical examination.

Findings at first examination: Three round tumors were observed. One was small, fingertip in size and was located with stalk on the anterior gingiva of the mandible; the other two on the right maxillary alveolar region were different in size (one being a red bean in size and the other being a soybean in size). These pedunculated tumors were firm, pinkish and smooth in surface. The mandibular tumor protruded beyond the lips, due to its relatively large size, but according to her mother it did

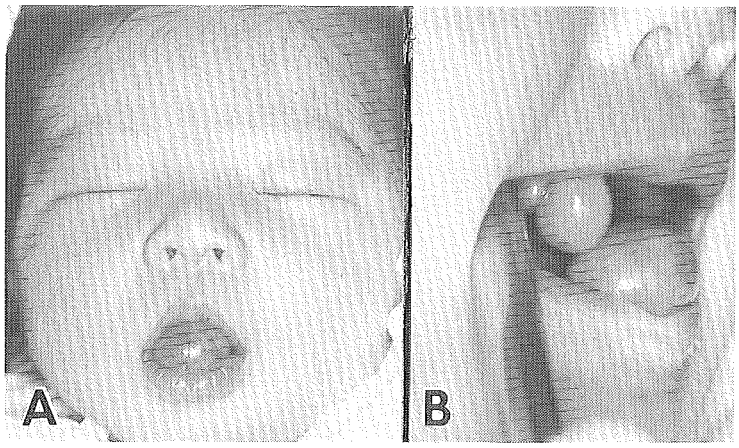

Fig. 1. Preoperative view of the multiple congenital epulides
A; Mandibular lump showing difficulty of the mouth closing due to its large size.
B; Positional relation of triple epulides

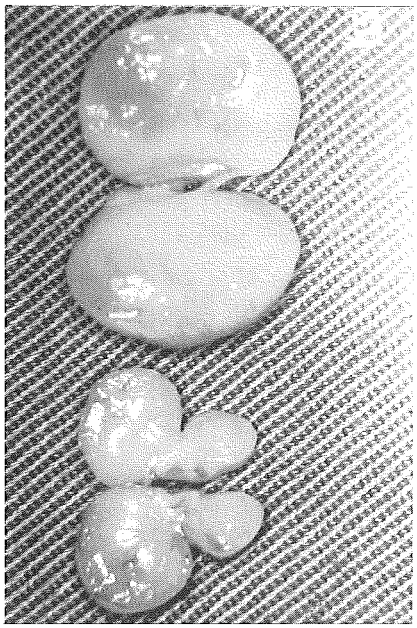

Fig. 2. Gross appearance of the cut surfaces

not interfere with her breathing and feeding (Fig. 1).

Clinical diagnosis: Probable congenital epulis.

Surgical procedure and time course: Under local anesthesia with Lidocaine, complete surgical excision of the lesions was performed. Exposed bone surfaces were normal. The postoperative course was uneventful and uncomplicated. Two years follow-up showed neither recurrence of the lesion nor abnormal course.

Gross findings: The surface of each tumor was smooth and round. The two tumors of the maxilla were partially connected at their bases. The cut surfaces of the three tumors were yellowish and homogeneous (Fig. 2).

Histopathologic findings: Each tumor consisted of large polygonal cells which were filled with eosinophilic fine granules and their nuclei were small. Almost no mitotic figures could be seen (Fig. 3). The overlying epithelia had become thin and flat, and groups of tumor cells were separated in places by fibrous connective tissues. In these connective

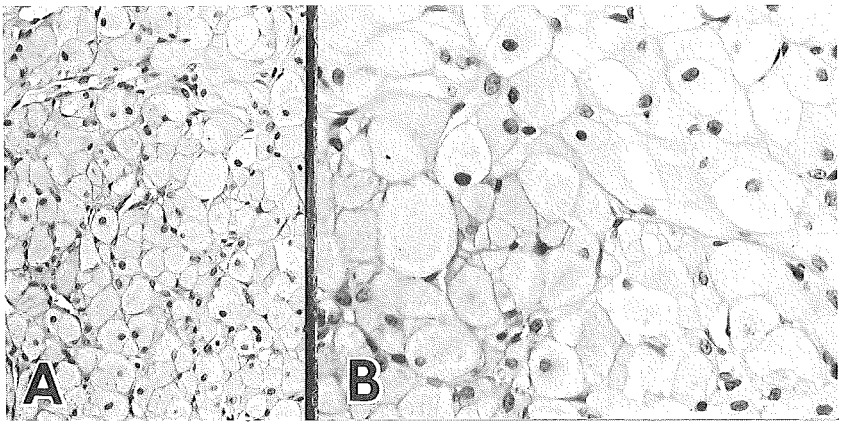

Fig. 3. Large polygonal cells with eosinophilic fine granules, which features the lesions (A; HE ×100, B; HE ×200)

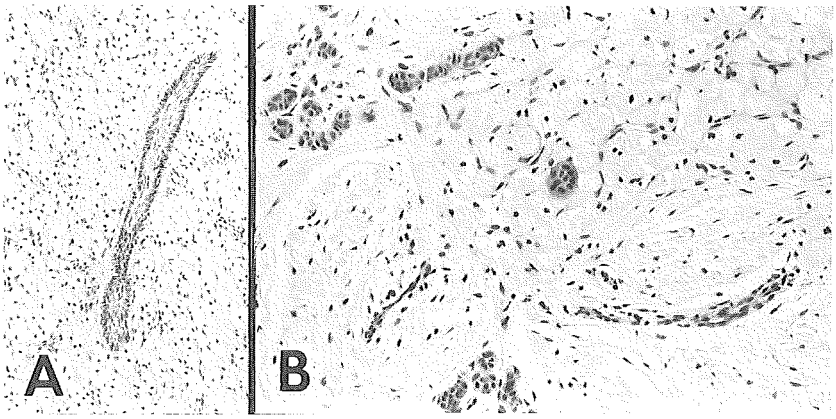

Fig. 4. Odontogenic cells adjacent to granular cells (A; HE ×50, B; HE ×100)

Table 1. Reports of congenital epulis in Japan

		Full papers	Abstracts	Total
Sex	female	14	16	30
	male	0	4	4
Location				
solitary	maxilla	10	5	15
	mandible	3	11	14
multiple	maxilla	0	1	1
	mandible	0	1	1
multiple	maxilla + mandible	1 (ours)	2	3

tissues among the granular cell nests, cell groups suggesting odontogenic origin could be seen in some parts (Fig. 4). The vascularities were also prominent.

Histological diagnosis: Congenital granular-cell epulis.

DISCUSSION

Webb et al[21] have reported that the incidence of congenital epulis in females is about nine times higher than that in males and that the occurrence in the maxilla is about two times greater than in the mandible. Most appear as a single lesion, but rare cases of multiple lesions have been reported[1,2,4,15,20]. It is a matter of interest that full papers on 14 cases[3,6-13,16,17,22] have been published in Japan, including the present case. All of these cases were famales. In ten cases, the lesion developed in the maxilla and in three cases in the mandible. Moreover, if abstracts are added, 34 cases have been reported in Japan in the past 54 years, as far as we know (Table 1). By sex, famales accounted for 30 cases and males for 4 cases. The occurrence of this lesion in females was about 7.5 times more frequent than that in males. The result shows that this tumor is prone to develop more in females than males also in Japan.

At present we have no hypotheses to account for female preponderance. The number of cases with single lesion in the maxilla was 15 and that in the mandible was 14. The ratio of incidence of the lesion in the maxilla to that in the mandible was almost the same in Japan, but in other countries it occurred in the maxilla three times more often than in the mandible[5]. In Japan, multiple lesion of the maxilla and of the mandible have been reported in one case, respectively (Table 1). To our knowledge, excluding the present case, two cases have been reported in which the lesion was observed both in the maxilla and the mandible. Similar cases have also been reported in other countries[1,15,20]. In our case, three tumors simultaneously occurred both in the maxilla and the mandible.

It is well known that cases of multiple lesions are accompanied by respiratory and feeding difficulties[1,4,15,20], but these can be resolved by surgical excisions. Fortunately, our patient did not have such disorders.

It is yet unknown whether congenital epulis is a true neoplasm or not and whether it is similar in etiology and entity to granular cell tumor of other sites. To date, the etiology of this lesion remains obscure and there are many hypotheses which suggest odontogenic, fibroblastic, histiocytic, myoblastic and/or neurogenic origins[5]. Many reserchers have suggested that these tumors are derived from malformed dental blastema and are embryonal hamartomas[18]. We would concur that the odontogenic components might play an important role in the occurrence of this lesion, because this lesion develops only on the gum of the newborn. Epithelial islets are frequently seen in normal fetal alveolar mucosa, and odontogenic-like cell nests among the granular cells are often observed as in this patient. In fact, Sunderland et al[19] have reported that granular cells are associated with the enamel organ of a developing tooth in a stillborn. This suggests the possibility that the lesion could develop from an enamel bud, resulting in a similarity with a congenital epulis. Furthermore, it is well known that there is a resemblance between granular cell ameloblastoma and congenital epulis. If the origin of granular cell is assumed to be the same as the degenerating changes found in granular cell ameloblastoma, there would be no recurrence without complete surgical excision and spontaneous regression will have taken place in some cases[4,16]. The nature and origin of this lesion cannot be fully explained only from these opinions. Further ultrastructural and immunohistochemical studies are needed in order to provide more comprehensive evidence.

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