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Congenital Epulis of the Newborn: Report of A Case with A Review of the Japanese Literature

Tsuneo Chiba and *Giichi Okayama

Department of Pediatric Surgery, Tohoku University School of Medicine, Sendai, Japan and
*Department of Surgery, Tsukidate Hospital, Miyagi, Japan
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Summary

Congenital granular cell tumor is a rare tumor which arises from alveolus. We describe a case diagnosed and treated in a neonate. Twenty-eight cases have been reported in Japan, with a male : female ratio of 1 : 6.3. The location of the tumor was the mandible in 13, the maxilla in 15 and multiple in 1. The left side incisor region was the most common site.

Introduction

Congenital epulis (congenital granular cell tumor) is a relatively rare tumor which has been reported as occurring most frequently in females[10]. A case report of congenital epulis is presented and the Japanese literature is reviewed.

Case Report

A girl weighing 3260 g, born by spontaneous vaginal delivery on June 12, 1989, was noted to have a fingertip-size tumor arising from the left maxillary alveolus (Fig. 1). She also had a large nevus on her right leg. There was no family history of congenital malformation. The mass was firm, whitish and fleshy with a partially ulcerated surface, measured $18 \times 14 \times 10$ mm, and had a pedicle, 8 mm in diameter. Though the tumor was not large, the infant nursed poorly.

The tumor was divided at its base by electrocautery and excised. Histological examination disclosed a surface of stratified squamous epithelium. The body of the tumor consisted of extremely large cells with marked granularity (Fig. 2). The cells contained a single, small, round nucleus. Many capillaries were interspersed throughout the tumor. The findings were consistent with a diagnosis of congenital granular cell tumor. Four hours after surgery, regular feedings were started. At the time of this report, there was no recurrence of the tumor.

Key words: Congenital epulis, Congenital granular cell tumor.
索引語：先天性エプーリス，先天性顆粒細胞腫.
Present address: Department of Pediatric Surgery, Tohoku University School of Medicine, 1-1 Seiryocho, Aobaku, Sendai Japan 980.

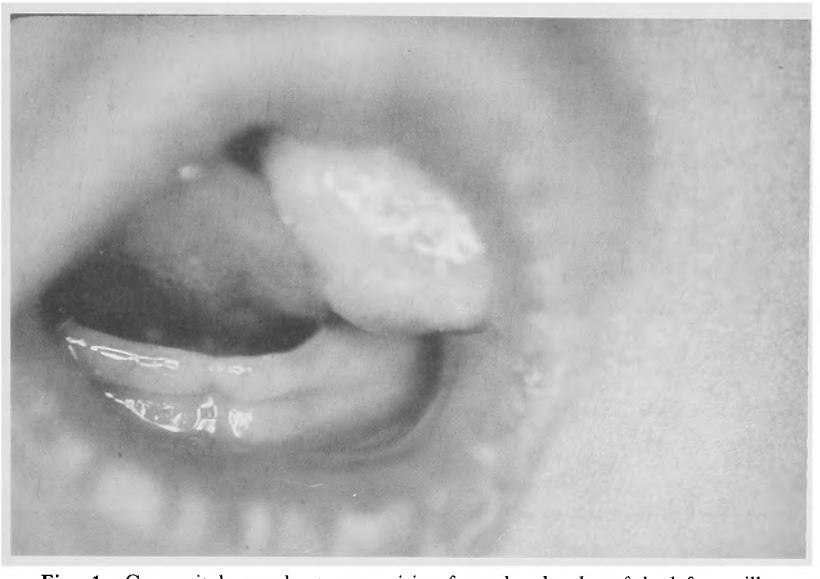

Fig. 1 Congenital granular tumor arising from the alveolus of the left maxilla.

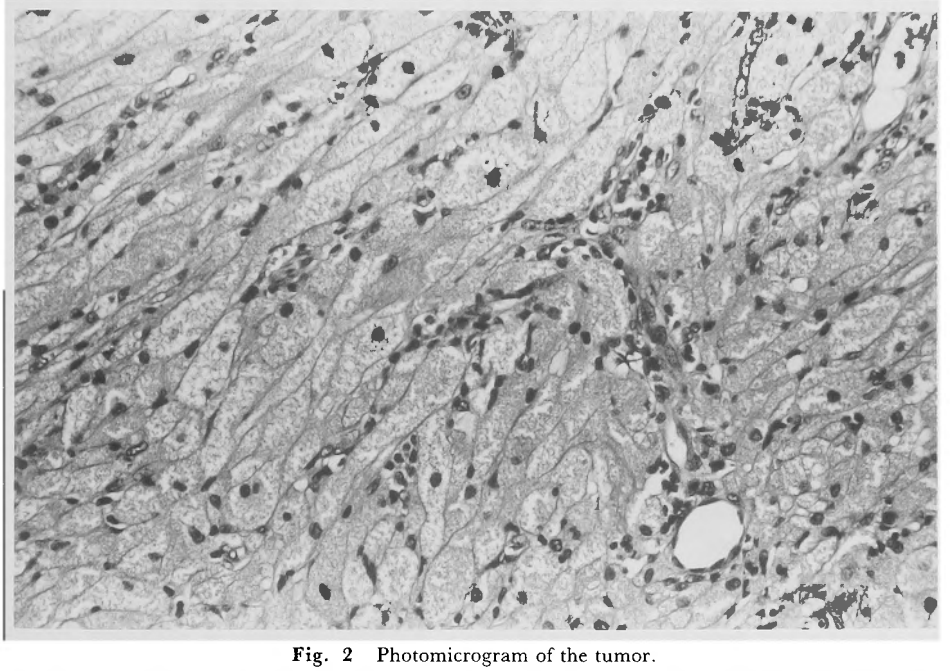

Fig. 2 Photomicrogram of the tumor.

Discussion

Congenital granular cell tumor (epulis) is a rare, benign tumor which usually arises from the alveolus. More than 120 cases have been reported in English since NEUMAN's first description in 1987[10]. This tumor occurs predominantly in females[3], and it develops two to four times more often in the maxilla than mandible[1,3,4] It may cause respiratory difficulty and nursing problems.

Tumors arising from the alveolus are not always granular cell tumors. Hamartoma and fibrous tissue must be considered, and the differential diagnosis is comparatively difficult based on clinical findings. As treatment, simple excision is recommended, although spontaneous regression also has been reported[11]. The prognosis is excellent and recurrence is extremely rare.

Twenty-eight cases of granular cell tumor have been reported in Japan since 1956 (Table 1)[2,5,6,7,9]. The male : female ratio was 1 : 6.3, and the tumor was on mandible in 13, the maxilla in 15 and multiple in 1 case. This pattern of localization is different from that in the English literature. It developed in the left incisor region in 15, the right in 6, central anterior in 6, bilaterally in 1 and not reported in 1 case. Its size ranged from 5 mm to 37 mm. Tumors in 23 patients were excised in the newborn period, and 5 others were excized after observation. There was one case of spontaneous sloughing. No tumor recurred.

Table 1 Reported cases of congenital granular cell tumor in Japan

Author	Year of report	Sex	Location	Size (mm)	Treatment
1 Kobayakawa[7]	1956	M	Mandible, central	Finger-tip size	?
2 Kimura[7]	1963	M	Mandible, right	Soybean size	surgery
3 Shimizu[7]	1968	F	Mandible, left	Soybean size	surgery
4 Eda[7]	1970	F	Maxilla, left	10	surgery
5 Sakamoto[7]	1970	F	Mandible, left	Soybean size	surgery
6 Masuda[7]	1971	F	Mandible, left	10×10×17	surgery
7 Takeda[7]	1973	F	Maxilla, central	22×18	surgery
8 Ohto[7]	1974	F	Maxilla, left	13×16	sloughed
9 Ito[7]	1976	F	Mandible, central	15×12×10	surgery
10 Hisaka[7]	1977	F	Mandible, right	12×12×10	surgery
11 Ishii[7]	1978	F	Maxilla, left	5×5×3	surgery
12 Yamada[7]	1979	F	Maxilla, left	12×7×7	surgery
13 Nagase[7]	1980	M	Maxilla, left	30×20×10	surgery
14 Matsui[7]	1980	F	Maxilla, left	15×19×10	surgery
15 Fukutake[9]	1980	M	Mandible, left	10×10	surgery
16 Kojimahara[9]	1981	F	Mandible, ?	Hen-egg size	surgery
17 Murata[9]	1981	F	Maxilla, left	7×3×3	surgery
18 Fujii[7]	1982	F	Maxilla, right	31×21×10	surgery
19 Kitano[9]	1982	F	Mandible, central	Thumb-tip size	surgery
20 Morita[7]	1983	F	Maxilla, right	25×15	surgery
21 Nakao[9]	1983	F	Maxilla, left	10×25	surgery
22 Kohno[6]	1984	F	Maxilla, central	37×29×20	surgery
23 Ikeda[5]	1984	F	Mandible, right	10×15×7	surgery
24 Ikeda[5]	1984	F	Maxilla, left	5×10×5	surgery
25 Endo[2]	1985	F	Mandible, right	15×10×5	surgery
26 Fukuya[9]	1985	F	multiple	5×6×16, 4×5×16	surgery
27 Nakatani[8]	1986	F	Maxilla, left	5	surgery
28 Nakayama[1]	1987	F	Mandible, central	8×6×6	surgery
29 Chiba		F	Maxilla, left	18×14×10	surgery

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和文抄録

新生児期先天性エプーリス
—本邦報告例の考察を加えて—

東北大学小児外科
千　葉　庸　夫
築館病院外科
岡　山　義　一

先天性エプーリスは歯齦部に発生する稀な疾患である．われわれは左上顎より発生した女児例を経験したので報告した．なお本邦における報告例28例を分析した．これによれば男女比は 1 : 6.3，部位は下顎13，上顎15，多発 1 であった．局在としては左切歯部が15例と最も多かった．新生児期に23例が切除されたが，自然消褪例が 1 例見られた．