Granular congenital cell tumor in the newborn: a case report

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This report represents a case of unusual large size congenital granular cell tumor appearing on the maxillary alveolar ridge in a newborn. Positive staining was found for S-100 protein. The authors discuss the clinical picture, histological findings, etiology and treatment of this rare lesion.
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INTRODUCTION

The most congenitally tumor like masses (termed congenital epulis CE, or congenital granular cell tumor CGCT) found in oral cavity of the alveolar crest of the newborn are predominantly single maxillary lesions, although some multiple lesion on both jaws have been reported.[1] Numerous theories of histogenesis were proposed. The most recent include epithelial, undifferentiated mesenchymal cells, pericites, fibroblasts, smooth muscles, nerve-related cells and myofibroblasts.[2] However, definite histogenesis remains controversial.

These lesions are very rare. They do not show active growth after birth and spontaneous regression occasionally been reported.[3] Although that the lesion is benign in its nature, it still can cause respiratory and feeding problems and considerably anxiety among parents.[4] Surgical excision is generally indicated and no recurrence has been reported.[5-17]

CASE REPORT

A one-day-old infant was referred to Oral and Maxillo-Facial Unit, Ministry of Health for the diagnosis and treatment of mass on the left side of maxillary alveolar ridge that was interfering with nursing and feeding the baby. The referring report described a well-developed newborn baby girl (body weight 4.425kg), born of normal and uncomplicated full-term pregnancy (from the couple father 43 years old and mother 36 years old). Intra and extra-oral examination revealed a soft tissue mass 4.0x3.5x2.0cm in diameter (Figure1), attached to the maxillary alveolar ridge of the left side at the canine area, of red color (Figure 2). The mass was elastic, ulcerated, pedunculated and freely movable. Medical signs were normal and there were no lymphadenopatphy.

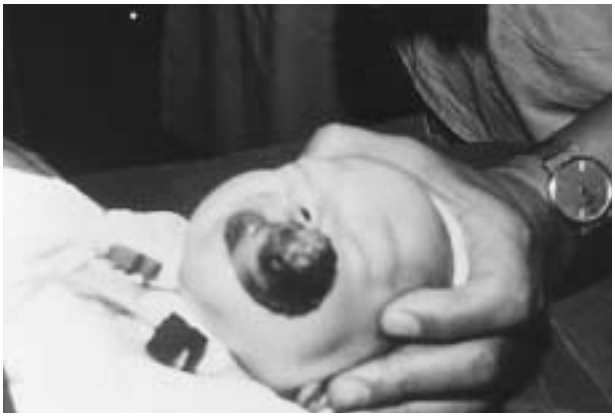

Figure 1. Clinical appearance of the tumor at birth.

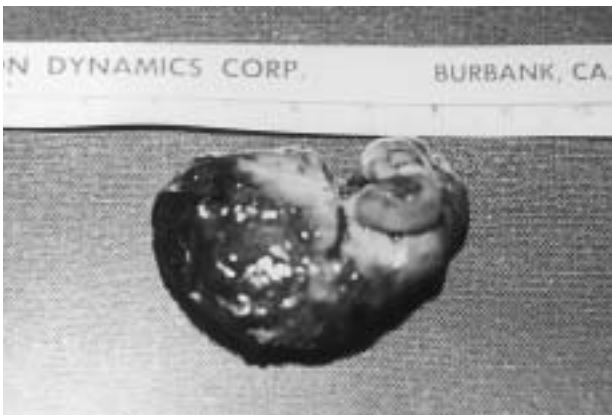

Figure 2. Macroscopical view of the tumor mass after excision.

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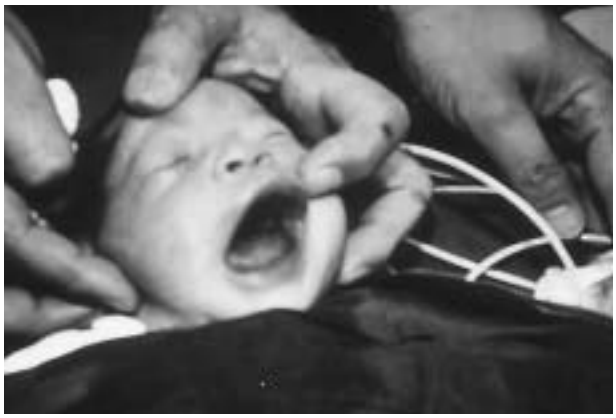

Figure 3. Postoperative view of the infant.

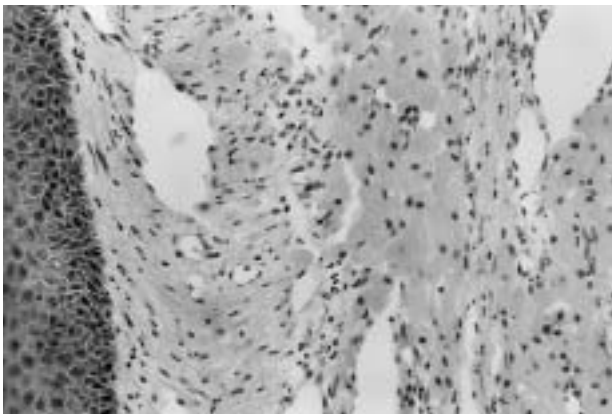

Figure 4. High power view showing basal cell layer without rete pigs. The underlying connective tissue is mainly fibrous with large vascular channels that lined by single layer of endothelium.

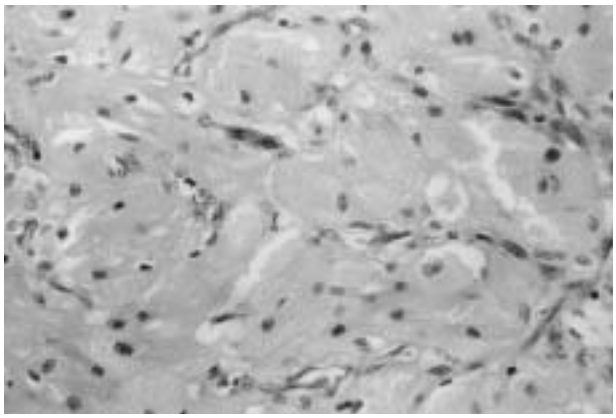

Figure 5. Section of the tumor showing cytoplasmic granules stained by period acid Schiff (PAS).

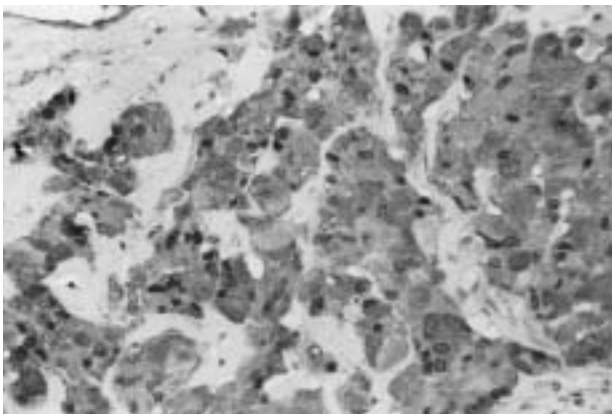

Figure 6. Section of the tumor cells stained positive for the S-100-protein using DAKO EPOS peroxidase reagent.

Surgical removal has been performed by simple excision between sutures with No. 15 Bard Parker scalpel blade. Before excision two ligation sutures were placed to the alveolar mucous as possible to prevent bleeding. Two vicryl stitches and application of pressure accomplished hemostasis over the region for 15 minutes. There were no adverse affects and infant was breast fed by her mother on the four day of birth (Figure 3).

Histopathological examination

The section showed non-keratinising stratified squamous cell mucosa ulcerated in areas overlying tumor of polygonal cells with granular eosinophil cytoplasm (Figure 4). The tumor cells were separated into nests by fibrovascular septa. The cytoplasmic granules were stained by period acid schiff (PAS) and were resistant to diastase digestion (Figure 5). The tumor cells stained positive for the S-100-protein using DAKO EPOS peroxidase reagent (Figure 6). Diagnosis was congenital granular cell tumor.

DISCUSSION

While the number of reported cases on congenital granular lesions of the newborn is appearing more in literature, even though it has a rare occurrence. The controversies continue regarding its terminology, histogenic origin and immunohistochemistry. At the present, granular cell tumor (myoblastoma, Abrikosoff's tumor) and congenital epulis in newborn (Neumann tumor) are regarded as identical pathological entity, but immunohistologicaly do not appear to be a homogenous group.[14] According to the same author sufficient data that support this hypothesis exist now days. Regarding GCT, most authors agree[9,14] that positive immunoreactivity for S-100 protein furnished strong evidence that this tumor is neural or neuroectodermal in origin. Beside invention of immunohistochemistry histogenic origin of epulis, still remain unclear and in most reported cases CE immunoreactivity to S-100 protein is negative.

It is an interesting finding, that our reported case has presented clinically almost identical as in size, shape

and location as a case presented by Jingo Kusukawa.[10] Immunohistologically we found S-100 protein positive reaction that was not in Jingo's reported case. Diagnosis made in Jingo reported case was congenital giant cell tumor on the basis of absence positive immunoreactivity to S-100 protein and Kp-1 (Cd68). The positivity for S-100 protein in our reported case collaborates with previous reports.[2,6,14,17] These data suggest that this tumor is slightly different from CE due to the difference in immunohistochemical reactivity to S-100 protein test.

Lindia[5] reported a case where a gingival mass in male infant appeared clinically as a congenital epulis, while histological examination revealed pyogenic granuloma. This illustrates the difficulty in achieving correct diagnosis of tumor like masses in newborn without histopathalogy. The importance of complete oral examination at the initial newborn visit has been suggested,[4,5] particularly in the light of advent of spectrum AIDS-related neoplasms that may occur at any age, and other lesions that may be aggressive. Treatment recommendation for all these tumors or tumor-like masses appearing at birth, is surgical removal. Recurrence and metastases have not been reported.

As Uglesic[11] mentioned, typical localization (anterior region of the maxillary alveolar crest), presents at birth, good vascularization, pedunculated growth, rare recurrence and no malignant alteration gives characteristics, which slightly differ from other granular cell tumor. In the same time analysis of more reported cases will contribute to a better understanding of the etiology and histogenesis of tumor masses appearing at birth in infants. An extensive battery of immunoperoxidase stains should be used in all cases to role out the possibility of origin.

The existing data are sufficient to make some researchers like Luis[14] to consider CE and GCT as two different entities, but still some controversy continues over terminology. In this article the use of the term CGCT was based on our pathological finding. Although, the term congenital epulis of the newborn suggested by Douglas[2] can be an appropriate term for this tumor mass due to its wide acceptance. It is important that new cases are reported from all populations so that the occurrence and frequency may easily noted.[4]

According to Peter[7] strong requirements are essential for the education of pediatric dentists in this field. The involvement of pediatric dentists in the teamwork on these cases will help them obtain the necessary skills to diagnose intraoral problems in newborn.

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