Congenital Fibrous Epulis in the Infant

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Congenital fibrous epulis is an extremely rare tumor of infancy. It is a benign gingival tumor and generally seen in maxillary alveolar crest and its etiology remains the subject of debate. Congenital fibrous epulis could be considered a hamartomatous lesion. Histologically it does not show the closely packed large granular cells necessary for the diagnosis of an ordinary congenital epulis. Instead, it consists of irregular bundles of collagenous connective tissue, varying numbers of fusiform cells with oval or fusiform shaped nuclei and mild subepithelial inflammatory infiltration with tiny blood vessels and in this case a woven bone spicule in the deep area. Recommended treatment for this tumor is simple excision. We report upon a case of congenital fibrous epulis in a male infant and discuss the clinical features, histopathologic findings, and surgical treatment.

Key Words: Congenital fibrous epulis, infant, maxillary gingival tumor

INTRODUCTION

Congenital epulis is an unusual benign oral cavity tumor that is present in the alveolar mucosa at birth. The tumor varies from 3 to 90 mm² is pedunculated, smooth surfaced¹² and arises from the alveolar mucosa.³ Congenital epulis is reported three times more frequently in the maxillary alveolus than in the mandibular alveolus and the female to male ratio is 10:1. In the etiology of the condition is not clear and is still argued, several theories have been suggested, namely, myoblastic, odontogenic, neurogenic, fibroblastic, histiocytic, and endocrinologic.⁴ Congenital fibrous epulis is a clinical and histopathologic variety of congenital epulis.¹² We were only able to find two reported cases in literature. In this paper, we report a case of congenital fibrous epulis in a male infant and discuss its clinical features, histopathologic findings and surgical treatment.

CASE REPORT

A five-month boy had a tumor in his gingival tissue. The patient’s history showed that his grandmother first noticed the tumor at birth and this was confirmed by his mother. The tumor did not appear to be interfering with feeding and respiration. There was no family history of trauma, chronic irritation or congenital abnormalities. On physical examination a soft tissue tumor was found attached to the right anterior maxillary alveolus. The tumor was fluctuant, smooth surfaced and red in color, much like alveolar mucosal tissue. It was well-circumscribed and 1 × 0.5 × 0.5 cm in diameter. Other clinical and radiological evaluations were normal. The possible diagnoses were epignatus or congenital epulis. The tumor was excised totally from its narrow pedicle by electrocautery under general anesthesia. Twenty hours after the operation regular oral feeding was started and was well tolerated. The surgical wound healed uneventfully and no recurrence was evident six months after operation.

The specimen was macroscopically pedunculated and smooth surfaced. Histologically, it was covered with a layer of stratified squamous epithelium showing irregular acanthosis and slight

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parakeratosis. The tumor was found to be composed of irregular bundles of collagenous connective tissue, varying numbers of fusiform cells with oval or fusiform shaped nuclei and mild subepithelial inflammatory infiltration with tiny blood vessels (Fig. 1). A woven bone spicule was found in the deep area. The lesion seemed to be composed of connective tissue and collagenous bundles by Masson’s trichrome and Verhoeff’s elastic stains. Neither the cells nor the collagenous bundles were immunopositive for desmin, S-100 protein, neuron specific enolase or Glial Fibrillary Acidic Protein. Various degrees of positive staining for vimentin were observed in several collagenous bundles and the cells embracing them. According to these findings, the case was interpreted as fibrous epulis.

**DISCUSSION**

Congenital fibrous epulis was first described by Majid et al and the second case was reported by Takeda et al. It is a regular, fluctuant, freely mobile, circumscribed gingival tumor at birth which in general originates from the alveolar crest of the maxilla. The average diameter of this tumor is approximately 9 mm and it is thus smaller than congenital epulis. It is a benign tumor and malignant transformation has not been reported. It should be differentiated from Epstein pearls, Bohn nodules, epignatus, pyogenic granuloma and congenital epulis (congenital gingival tumor).

Although congenital fibrous epulis is claimed to be identical with congenital epulis and to involve predominately female infants, the three cases (including the present case) in the literature concern one female and two male babies. It is well known that congenital epulis does not grow after birth and sometimes undergo spontaneous resolution. However, the present case showed gradual growth. The patient was evaluated initially at five-months when the tumor had already grown. Histologically it did not show the closely packed large granular cells necessary for the diagnosis of an ordinary congenital epulis. Therefore, we believe that congenital fibrous epulis can be considered as a distinct histological and clinical entity.

Fibrous epulis in adults occurs due to chronic irritation or trauma, due to factors such as dentures, a carious tooth, faulty restorations, and subgingival calculus. In the present case there was nothing to explain inflammatory reaction. However, histologically a woven bone spicule, was present in deep area which has previously been seen in the fibrous epulis of the adult. We suggest that congenital fibrous epulis is a benign hamartomatous tumor and that it differs from other congenital fibrous proliferative lesions seen in gingiva. This tumor is neither congenital epulis nor fibrous epulis and should be accepted as a special tumor and called congenital fibrous epulis.

We believe that recurrence will not occur if the lesion is excised totally. Early surgical excision may be performed to prevent airway obstruction and feeding difficulties. Extensive surgery is unnecessary and can damage underlying unerupted dentition. We prefer simple excision by electrocautery under general anestheisia. Resection by carbondioxide laser may be another surgical

*Fig. 1. Congenital gingival tumor under light microsceopy. The lesion consists of fibrovascular tissue with keratin forming stratified squamous epithelium with irregular acanthosis and slight parakeratosis The fibrovascular tissue consisted of fibroblasts as a shoal of fish tiny blood vessels with collagen bundles and a woven bone spicule in the deep area (H+E, × 100).*
treatment option.6

REFERENCES