

## Case Report

# Congenital fibrous epulis associated with natal teeth

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**Abstract:** Congenital fibrous epulis is a very rare benign gingival tumor in infants. Its exact etiology remains unknown. The past literature showed that there was no other abnormality around or in the tumors in their mouths. In our clinic we found seven cases of congenital fibrous epulis and all the cases had one or two natal teeth in the tumors. We described the clinical and histopathologic characteristics of the lesions, treatments and discussed whether congenital fibrous epulis was related to natal teeth. Natal teeth may be probed as a causative factor of congenital fibrous epulis.

**Keywords:** Congenital fibrous epulis, natal teeth, infant, oral surgery

### Introduction

Congenital fibrous epulis is a very rare benign gingival tumor in infants and was first described by Majid et al in 1986 [1]. To our knowledge, there are only four cases reported in the past literature in English [1-4]. Its exact etiology remains unknown. Majid et al [1] thought that congenital fibrous epulis could be considered as a hamartomatous entity since it developed in utero. According to Inan et al [3], congenital fibrous epulis is neither congenital epulis nor fibrous epulis, and can be considered as a distinct histological and clinical entity. All the reported cases showed that the epulis were first noticed at birth and there was no other abnormality around or in the epulis in their mouths.

Natal teeth in humans are rare and often erupt in the mandible, especially affecting the lower primary central incisors. Less than 10% natal teeth are supernumerary. Natal teeth can bring many problems including discomfort during sucking, laceration of mother's breasts, sublingual ulcer and aspiration of the teeth [5]. There were some reports about natal tooth associated with soft tissues such as peripheral ossifying fibroma [6], pyogenic granuloma [7], eruption cyst [8]. But in the past literature published in English and Chinese there was no any report about congenital fibrous epulis associated with

natal teeth. There was a report about congenital epulis with natal tooth published in Japanese [9]. In this report the histopathological examination showed that the epulis consisted of fibrous connective tissue covered by layers of stratified squamous epithelium. However, the diagnosis was congenital epulis.

In our clinic we found seven cases of congenital fibrous epulis and all the cases had one or two natal teeth in the masses. In this report we described their clinical findings, histopathologic findings, treatments and discussed whether congenital fibrous epulis was related to natal teeth.

### Case presentation

From February 2009 to August 2012, seven babies (5 males and 2 females) were referred to our clinic for the masses in their gingival tissue (**Table 1**). The age ranged from 1M29D to 25M5D. All the masses were first noticed at birth and one or two teeth were found in the mass. We presented 2 typical cases: one mass located in the mandible and the other located in the maxilla.

#### Case 1

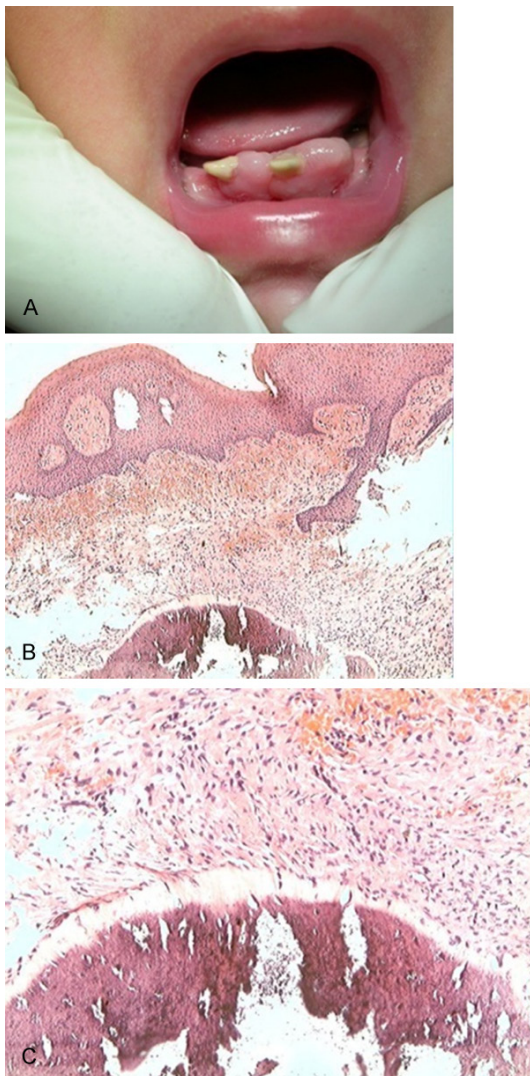
A 7 months and 5 days old male infant was referred to our clinic with two masses in his

## Congenital fibrous epulis and natal teeth

**Table 1.** Summary of the seven cases in our study

Case	Age	Gender	Natal teeth and lesion site	Lesion size	Chief complaints	Treatment	Recurrences (follow-up period)
1	7M5D	M	71, 81 Mandible	1.5*2.5*0.8 cm; 1.5*1.5*0.8 cm	Feeding interference	EE	None (24 M)
2	25M5D	M	Supernumerary Maxilla	1.0*0.8*0.5 cm	Feeding interference	EE	None (18 M)
3	5M15D	M	Supernumerary Maxilla	1.2*0.8*0.5 cm	Feeding interference	EE	None (12 M)
4	4M15D	M	71 (dropped) Mandible	1.0*0.8*0.6 cm	Feeding interference	Excise the tumor	None (12 M)
5	1M29D	F	71 Mandible	1.0*0.5*0.5 cm	Feeding interference	EE	None (18 M)
6	5M3D	F	71 Mandible	1.0*0.8*0.5 cm	Feeding interference	EE	None (35 M)
7	7M3D	M	71 (dropped) Mandible	0.8*0.5*0.6 cm	Feeding interference	Excise the tumor	None (36 M)

Note: EE: extract the teeth and excise the tumor.

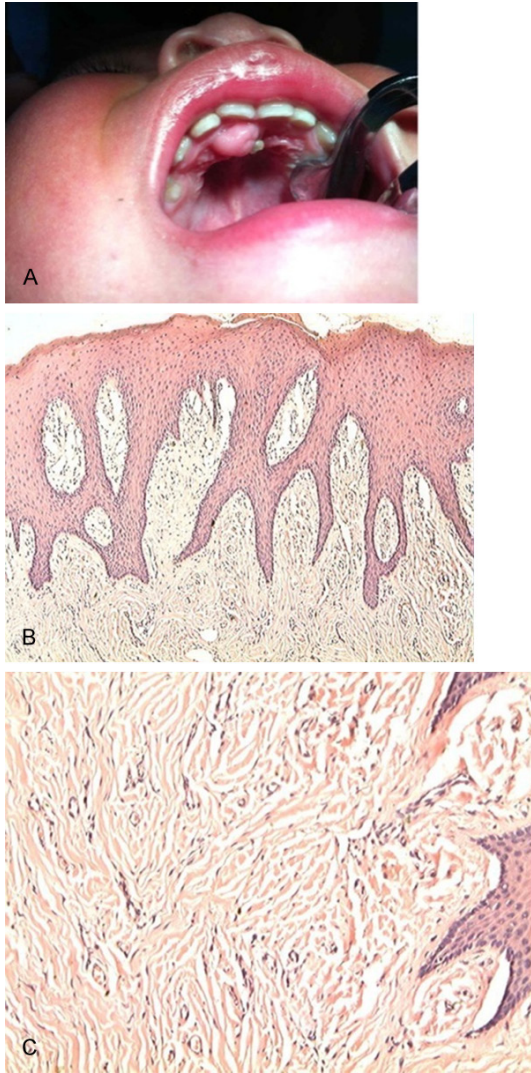


**Figure 1.** A. Clinic appearance of case 1: two tumors attached to the top of the anterior mandibular alveolus and two natal teeth (71, 81) in the tumors. B. Histopathologic characteristics of case 1 (H-E,  $\times 40$ ). C. Histopathologic characteristics of case 1 (H-E,  $\times 100$ ).

mandibular gingival tissue (**Figure 1A**). Two masses were noticed at birth in the middle of the mandible and in which there were two teeth. The masses showed gradual growth and resulted in feeding difficulties. Physical examination revealed that there were 2 soft tissue masses attached to the top of the anterior mandibular alveolus and 2 teeth with a mobility of 2-3 degree in the masses. The position of the 2 teeth was 71 and 81. The masses were fluctuant, pedunculated, smooth surfaced and covered with normal pink mucosa. The sizes of the masses were about 1.5 cm\*2.5 cm\*0.8 cm and 1.5 cm\*1.5 cm\*0.8 cm and the width of the pedicle was about 0.3 cm. The crown size, shape and color of the teeth were normal. There was no family history of congenital abnormality and the child was otherwise normal. The primary diagnoses were natal teeth and congenital epulis. Under local anesthesia the two teeth were extracted. The roots of the teeth were poor formed. After two weeks followed up, there was no any change in the size of the masses. The masses were excised totally from their narrow pedicles under local anesthesia, and then were sent for histopathological examination. The surgical wound healed uneventfully and in the later 24-month follow up, there was no evident recurrence.

Histological examinations revealed that the masses were composed of the proliferated fibrous connective tissue and ossified partly, in which there were bundles of collagen fibers (**Figure 1B, 1C**). Inflammatory cells and small veins were seen in the connective tissue. The lesion tissue was covered with a stratified squamous epithelium. There were no any closely packed large granular cells. The histological appearance was consistent with a fibrous epu-

## Congenital fibrous epulis and natal teeth



**Figure 2.** A. Clinical appearance of case 2: a tumor about 1.0 cm in diameter on the maxilla, just at the palatal side of the 51 tooth and with one tooth in the tumor. B. Histopathologic characteristics of case 2 (H-E,  $\times 40$ ). C. Histopathologic characteristics of case 2 (H-E,  $\times 100$ ).

lis. On account of these findings, the case was diagnosed as congenital fibrous epulis.

### Case 2

A 25 months and 5 days old male baby had a mass in the maxilla, just at the palatal side of the 51 tooth (**Figure 2A**). The mass was about 1.0 cm\*0.8 cm\*0.5 cm in diameter with a tooth. The mass was a fluctuant mass covered with normal pink mucosa. The parent's history showed that at birth there was a small mass at the palatal side of the anterior maxilla with a loose, conical and yellowish tooth and the mass

grew gradually. The mass caused feeding interference. The tentative diagnoses were natal teeth (supernumerary) and congenital epulis. Under local anesthesia the tooth was extracted and the mass was excised. The root of the extracted tooth was devoid. Histopathologic findings showed the mass was composed of proliferated fibrous connective tissue densely packed mature bundles of collagen fibers. The lesion tissue was covered with a stratified squamous epithelium with rete pegs extended. There were very few inflammatory cells and tiny blood veins in the connective tissue. However there were no any closely packed large granular cells (**Figure 2B, 2C**). Pathologic diagnosis was congenital fibrous epulis.

### Discussion

Congenital fibrous epulis is an extremely rare tumor of infancy and often shows a fluctuant, mobile mass covered with normal pink mucosa attached to the alveolar crest at birth in the oral cavity. It is often misdiagnosed before surgery. The differential diagnosis includes congenital epulis, peripheral ossifying fibroma, Epstein pearls and Granuloma [3, 10]. **Table 2** shows that congenital fibrous epulis reported in English literature often occurs in male infants (male/female ratio: 3:1) and is more frequently located in the maxilla with a ratio of 3:1. The masses can cause feeding interference and bleed during feeding. But all the cases showed that there was no other abnormality around or in the masses. Our report is unusual case with one or two natal teeth in the masses.

Congenital fibrous epulis is so rare that is lack of awareness by clinician. It should be differentiated from congenital epulis. Congenital epulis was first described by Neumann in 1871 and is known as congenital gingival granular cell tumor or Neumann's tumor. It often occurs on the gingival mucosa of the anterior alveolar ridge of the maxilla or mandible (maxillary/mandibular ratio 3:1) and the female suffer more frequently than the male (female/male ratio 8:1) [11]. **Table 1** shows the ratio of sex about congenital fibrous epulis is 5:2 (male:female) in our report, which is similar to the previous reports about congenital fibrous epulis, but is quite different from female predilection congenital epulis. While the maxillary/mandibular ratio that congenital fibrous epulis occurred in our report is 2:5 which is different from the

# Congenital fibrous epulis and natal teeth

**Table 2.** Cases of congenital fibrous epulis reported in English

Author	Age	Gender	Lesion size	Lesion site	Chief complaints
Majid et al (1986) [1]	2 W	M	1.5 cm*1.0 cm*1.5 cm	Maxilla	The tumor was bleeding and baby was fretful during feeding
Takeda et al (1990) [2]	14 D	F	4 mm in diameter	Maxilla	None
Inan et al (2002) [3]	5 M	M	1 cm*0.5 cm*0.5 cm	Maxilla	None
Manjunatha & Das (2009) [4]	4 M	M	2.0 cm*1.0 cm*1.5 cm	Mandible	Feeding interference

past literature. That might be due to natal teeth are often erupted in the mandible, especially affecting the lower primary central incisors. After birth congenital epulis does not grow and sometimes undergo spontaneous regression [12-15]. However congenital fibrous epulis may grow gradually, all the cases in our study showed gradual growth. The histopathologic findings showed that the masses were composed of proliferated fibrous connective tissue densely packed mature bundles of collagen fibers and were covered with a stratified squamous epithelium. Closely packed large granular cells could not be found in the histopathologic examination which is necessary for the diagnosis of congenital epulis [10]. Thus the masses in our report should be diagnosed as congenital fibrous epulis rather than congenital epulis.

Most of the natal teeth are mobile and can bring many problems. Whether natal teeth could cause any epulis was very rarely reported. Akizuki et al [9] reported a case of congenital epulis with natal tooth in a 6-month-old girl. In the report histopathological finding showed the epulis consisted of fibrous connective tissue covered by layers of stratified squamous epithelium. Kohli et al [6] reported that a two-hour-old female infant suffered from a cyst-like mass in the anterior mandibular ridge area at birth and a neonatal tooth erupted after one week. The mass was diagnosed as a peripheral ossifying fibroma by histological examination. Histological examination in Kohli et al's report revealed a lesion consisting primarily of granulation tissue with an ulcerated surface. In the connective tissue there were areas of dystrophic calcification and osteogenesis.

The pathogenesis of congenital fibrous epulis is still unclear. Congenital fibrous epulis is a histological variety of congenital epulis that can be considered as a hamartomatous entity by Majid et al [1]. The patients' histories of the past reported four babies showed that there was no abnormality in their oral cavity. But the seven

cases in our report showed all the patients had one or two natal teeth in the mass, except that two patients' natal teeth dropped before they were referred to our clinic. Several etiologic factors such as chronic irritation, trauma, dentures, a carious tooth, faulty restorations, and subgingival calculus have been suggested for the incidence of fibrous epulis in adults [16]. In our present report, all the masses packed one or two natal teeth, so we deduced that congenital fibrous epulis might result from the inflammatory reaction of the loose natal teeth.

The treatment for congenital fibrous epulis is excision under general or local anesthesia. Early surgical excision is performed to prevent feeding difficulties and airway obstruction. But extensive resection is unnecessary because it may damage the underlying unerupted dentition [3, 17]. In our report, all the masses were excised under local anesthesia. No complications and recurrences occurred during 12-36 months follow-up. Removal of natal teeth is indicated when they are either supernumerary, poorly-developed, feeding interference, or excessively mobile, which is associated with a risk of aspiration [18]. In this paper, five patients' natal teeth were extracted along with the masses except two patients because their natal teeth had dropped before they came to our clinic.

According to the limited case report, we concluded that congenital fibrous epulis associated with natal teeth has never been reported in the past literature. Natal teeth may be probed as a causative factor of congenital fibrous epulis. However the relationship between the congenital fibrous epulis and the natal teeth need a further study.

### Disclosure conflict of interest

None.

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## Congenital fibrous epulis and natal teeth

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