

Dental Management of Congenital Granular Cell Lesion and Neonatal Teeth: A Case Report

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Congenital granular cell lesion (CGCL) is a rare benign oral cavity tumor in infants. Neonatal teeth are also rare dental anomalies that appear during the first month of life. This report describes a case of eruption of neonatal teeth after surgical excision of CGCL. Surprisingly, residual neonatal teeth erupted after extraction of the neonatal teeth. If neonatal teeth are mobile, they should be carefully extracted with curettage of the underlying tissues of the dental papilla; failure to curette the socket might result in eruption of odontogenic remnants. If neonatal teeth were exfoliated, parents should be informed of the need for regular checkups with a dentist due to possibility of development of residual neonatal teeth.

Keywords: Congenital granular cell lesion, Neonatal teeth, Residual Neonatal teeth, Management

INTRODUCTION

Congenital granular cell lesion (CGCL) of the newborn, also known as congenital epulis, is a rare benign tumor of the oral cavity that was first identified by Neumann in 1871.¹ The tumor has been found to be 8 to 10 times more common in females than in males.^{2,3} It is generally a 1 to 2 cm diameter protuberant mass, although tumors up to 9 cm have been described. CGCL can be found as a solitary nodule or as multiple nodules.^{2,3} This tumor arises from the mucosa of the gingiva, most commonly from the anterior part of the maxillary alveolar ridge. However, occasional cases have also been reported on the mandibular alveolar ridge or tongue.^{3,4} The lesion often presents as raised smooth pink mucosa but may also be erythematous or ulcerated. Bone or dental anomalies are usually not present.⁴ CGCL can interfere with both feeding and breathing of the newborn.⁵ Surgical excision of the lesion is generally indicated, and recurrences have not been reported. In some cases of small CGCL, spontaneous regression has been reported.^{6,7}

Natal teeth are defined as those present at birth, and neonatal teeth are those that erupt during the first month of life;^{8,9} the incidence varies from 1:1,000 to 30,000.⁸ The most commonly affected teeth are the lower primary central incisors.^{8,9} As a general rule, natal and neonatal teeth should be retained because they are most frequently teeth of normal primary dentition. The risk of consequent aspiration, in addition to traumatic injury to the baby's tongue and/or to the maternal breast, have been described as reasons for removal.¹⁰

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Until recently, there were no reports about CGCL associated with natal or neonatal teeth. Moreover, complications after extraction of natal or neonatal teeth have been reported only in a few cases.¹¹⁻¹⁶ The purpose of this case report is to describe tooth-like structures that developed after extraction of neonatal teeth following excision of CGCL and to suggest how to manage such occurrences in infants.

Case report

A 12-day-old girl was referred to Jeonbuk National University Dental Hospital for gingival overgrowths that were causing feeding problems. On clinical examination, two soft tissue masses were attached to the mandibular anterior alveolar ridge. The masses were fluctuant, pedunculated, and covered with pink mucosa. Two soft tissue lesions on the tongue were also observed (Fig. 1A). The lesions were completely excised under general anesthesia (Fig. 1B). Histologically, the lesions were attenuated overlying squamous epithelium with granular cytoplasm among vascular proliferation (Fig. 1C). Immunostaining for vimentin and α 1-antitrypsin was positive, while S-100, SMA, and cytokeratin immunohistochemical stains were negative. The final diagnosis was CGCL.

One week after surgery, the patient’s parents reported erupted teeth (Fig. 2A) in the anterior mandibular region. The crown size,

shape, and color of the teeth were normal. The teeth, diagnosed as “neonatal teeth” because they erupted within the first month of life, displayed severe mobility, causing risk of aspiration. Those were extracted simply (Fig. 2B), and curettage of the extraction site was not performed.

Three years later, the patient was again referred to Jeonbuk National University Dental Hospital by her family dentist. Tooth-like structures had erupted on the anterior mandibular alveolar ridge (Fig. 3A). Crown parts of the tooth-like structures were cone shaped and yellowish in color. On radiographic examination, there were periapical inflammatory lesions of tooth-like structures (Fig. 3B). Under local anesthesia, the structures were extracted, and curettage of the area was performed. Those structures were analyzed with radiographic micro-computed tomography (Fig. 3C, 3D) and were shown to have root canal structures with long axis. They showed uniform radiolucency and were missing the enamel layer of the tooth.

There were no clinical signs at the 1 year follow up (Fig. 4A). Radiographic examination revealed no remarkable pathologic signs and normal eruption of the permanent mandibular central incisors (Fig. 4B).

Figure 1. (A) Intraoral image of a 12-day-old infant with congenital granular cell lesion on the anterior mandibular alveolar ridge and tongue **(B)** Excised pedunculated tumors (Right lesion size : 1.5 x 1.0 cm, Left lesion size : 1.0 x 0.5 cm, tongue lesions : 0.2 to 0.5 cm) **(C)** Microscopic examination of excised specimen overlying squamous epithelium with granular cytoplasm (H&E stain, x100).



Figure 2. (A) Postoperative clinical image showing eruption of neonatal teeth on anterior mandibular area. **(B)** Extracted neonatal teeth with only crown part.

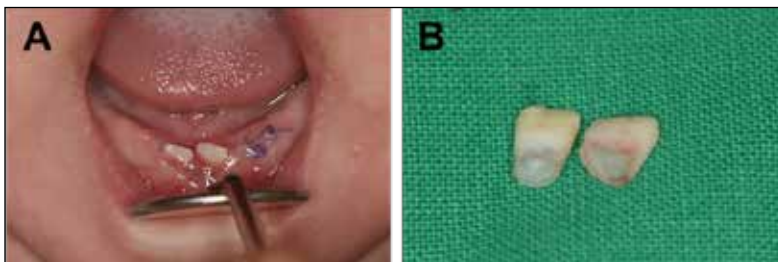


Figure 3. (A) Intraoral image of residual neonatal teeth showing yellowish color and cone-shaped. **(B)** Periapical radiograph of tooth-like structures showing periapical inflammatory lesion. **(C)** Micro CT image on #81 showing root canal structure and lack of coronal part. **(D)** Micro CT image on #71 also showing root canal structure with partial destruction on crown part.

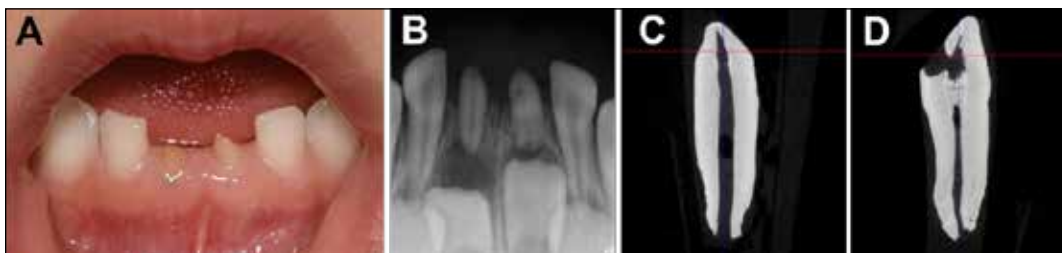
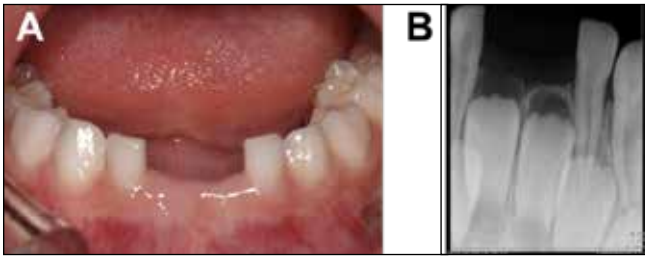


Figure 4. (A) Intraoral image of follow up check. No clinical signs on extraction site of #71, 81 area. (B) Periapical radiograph showing no remarkable pathologic sign.



DISCUSSION

CGCL of the newborn is a rare benign soft tissue tumor that usually arises from the anterior part of the maxilla.^{2,3} The tumors can appear in infants (0 to 1 year of age). Differential diagnosis should include granular cell tumors (GCT), fibromas, hemangiomas, lymphangiomas, granulomas, epignathus and gingival cysts.¹⁷ Diagnosis of CGCL is confirmed histologically; microscopic features of CGCL include granular cells, attenuated overlying squamous epithelium, and a capillary network. Immunohistochemically, the tumors lack S-100 staining. CGCL typically lacks immunoreactivity for S-100, while positivity is noted for vimentin and neuron-specific enolase. Due to similarity of clinicopathologic characteristics, CGCL and adult GCT were once thought to be closely related. In most cases, CGCL is distinguished from GCT by lack of S-100 immunoreactivity. Based on this lack of reactivity, the lesion is assumed to be derived from a different cell origin than conventional GCT in adults.^{4, 17, 18} The histological features of this case confirmed the lesion to be consistent with a CGCL.

The recommended treatment for large CGCL or multiple CGCL is surgical excision under either local or general anesthesia.^{4, 17} In some cases, if the patient had no airway obstruction or feeding problems, the lesion can be allowed to spontaneously regress.^{6, 19} Despite the aggressive appearance of CGCL, it does not invade alveolar bone. There have not been previous reports of recurrence when remnants remain following excision of the lesion.⁵ In the case presented here, there were CGCL, neither of which were obstructing the airway; however, because they were causing a feeding problem, surgical excision was performed under general anesthesia.

CGCL has an excellent prognosis. Residual remnants of the lesion have not been found to interfere with subsequent tooth eruption.^{4, 5} However, in the case presented here, after surgical excision of CGCL, neonatal teeth erupted. One study reported that congenital fibrous epulis was associated with natal teeth.²⁰ Congenital fibrous epulis is distinguished from CGCL and can be considered as a distinct clinical and histologic features. Clinically, CGCL do not grow and sometimes undergo spontaneous regression after birth; however, congenital fibrous epulis may grow gradually. The histopathologic findings showed that congenital fibrous epulis are composed of proliferated fibrous connective tissue and densely packed mature bundles of collagen fibers without the presence of large granular cells. CGCL associated with natal or neonatal teeth has not been reported in the literature. Considering that CGCL occurs predominantly in the maxillary anterior region, if the lesion is observed in the mandibular anterior region, it could be associated with natal or neonatal teeth. Therefore, conservative treatment is

recommended to preserve unerupted teeth buds if the patient has no respiratory or feeding problem.

There have been some case reports of complications following extraction of natal or neonatal teeth. Some pathological lesions, including tooth-like structures, fibrous haematoma, pulp polyp, continued growth of dental papilla, and peripheral ossifying fibroma, have been reported.¹¹⁻¹⁶ The phrase “residual neonatal teeth,” which has been used in some case reports, describes the tooth-like structures after extraction of neonatal teeth.^{11, 13} Because similar structures were presented in this case, we used the term in this report.

A previous report noted that stem cells from the pulp of human exfoliated deciduous teeth are capable of differentiating into odontoblast- and osteoblast-like cells and forming dentin and bone.²¹ Another study reported that mesenchymal stem cells (MSCs) from apical papilla were able to differentiate into dentinogenic cells.²² MSCs are capable of forming odontoblast-like cells that produce dentin and are likely to be the cell source of primary odontoblasts for formation of root dentin.²³ After formation of the tooth crown is nearly complete, the root develops with the guidance of the outer and inner layers of the epithelial sheath, called the Hertwig’s epithelial root sheath (HERS). The HERS is formed from two layers of enamel epithelium at the cervical region of the tooth crown and grows apically.²⁴ In this case, residual neonatal teeth consisting of dentin and pulp erupted 3 years after extraction of neonatal teeth. Enamel was not detected. One hypothesis for development of residual neonatal teeth after extraction of neonatal teeth is that the crown part may be exfoliated, while apical dental tissues including dental papilla and HERS remain and continue to develop, ultimately erupting as neonatal teeth. Signaling from HERS to adjacently undifferentiated MSCs encourages them to transform into odontoblast-like cells that produce residual neonatal teeth.

Regarding management of natal and neonatal teeth, there is an agreement among clinical scientists that natal and neonatal teeth should be retained because they mostly consist of normal primary dentition. An exception is made for teeth that appear to be excessively mobile and should be extracted because there is a possibility of swallowing or aspiration.¹⁰ Interestingly, however, not all clinicians have the same guidelines for extraction of natal and neonatal teeth. Some researchers suggested that, after extraction, curettage of underlying tissues of the dental papilla is necessary to prevent residual neonatal teeth. For such curettage, they recommend local anesthesia.^{15, 25-27} On the other hand, some authors recommend no curettage of the extraction site because risk of residual tooth formation is very rare.^{13, 28} The frequency of residual tooth eruption is not well established, and there is only one published report that suggests that 9.1% of infants with natal or neonatal teeth developed residual teeth following exfoliation or extraction of those teeth;²⁵ this suggests that formation of residual neonatal teeth is very rare following extraction of natal or neonatal teeth, which is also rare. Since curettage of the socket requires use of local anesthesia, which has a risk of inducing dental trauma, it is considered an aggressive approach. It is possible to remove natal or neonatal teeth with only topical anesthesia, and curettage with local anesthesia should only be performed in certain cases. However, considering that a risk of residual tooth formation is about 9.1%, dentists must inform parents about the need for regular checkups, and they should also be informed that, in case of residual tooth formation, an additional surgical procedure may be required.

CONCLUSION

In this case, CGCL occurred in the mandibular anterior region, and excision of those lesions affected the neonatal teeth. If neonatal teeth are mobile, they should be carefully extracted with curettage of the underlying tissues of the dental papilla; failure to curette the socket might result in eruption of odontogenic remnants. If neonatal teeth were exfoliated, parents should be informed of the need for regular checkups with a dentist due to possibility of development of residual neonatal teeth.

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