From Newborn to Toddler: Report of Two Cases of Congenital Granular Cell Tumor

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A congenital granular cell tumor (CGCT) is an uncommon benign soft tissue lesion that usually arises from the alveolar ridge in newborns. It can severely interfere with the respiratory and feeding systems, if left untreated. We present the cases of 2 newborn infants with protruded intraoral tumors that severely compromised breastfeeding. The tumors were removed by water laser without the need for suturing or pain medication. Histopathologic assessment was performed for definitive diagnosis, and the children were followed until full primary dentition was observed without any sign of CGCT recurrence.

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Congenital granular cell tumor (CGCT), also known as congenital epulis, congenital granular cell lesion, congenital myoblastoma, or Neumann's tumor, is a rare benign soft tissue tumor[1] that appears exclusively in newborns. Initially described by Neumann in 1871,[2] CGCT is usually present at birth as a single tumor; however, 10% of the cases will be multiple lesions with the size varying from a few millimeters to several centimeters.[3] The CGCT physically appears alarming owing to its large size and aggressive appearance, and it can physiologically interfere with the child's breathing and feeding.[1,2]

CGCT is usually located in the premaxillary region, with a predominant frequency in the maxilla (maxilla/mandible ratio 3:1)[1] and among females (female/male ratio 8 to 10:1).[4] The etiology of CGCT is unknown; however, maternal hormonal influence has been suggested owing to the predominance in females and the cessation of its growth or even spontaneous regression after birth.[5] The treatment option for large epulis has usually been simple surgical excision with the patient under either local or general anesthesia.[6] Despite the aggressive appearance of the CGCT, this tumor does not invade local bone. Also, in cases in which the remnants remained after excisional biopsy, no recurrences have been reported.[7] Histologically, CGCT is similar to the granular cell tumor; however, the overlying stratified squamous epithelium in CGCT does not show pseudoepitheliomatous hyperplasia. Distinctively, the microscopic specimen will show large round cells with abundant eosinophilic granular cytoplasm.[4-6] The present study reports the diagnosis, histopathologic findings, treatment, and prognosis of 2 patients with CGCT and their follow-up until full primary dentition had occurred at Kyung Hee University, Dental Hospital (Seoul, Korea).

Case Report

CASE 1

An otherwise healthy male baby, delivered at a local obstetrics hospital, was referred to our pediatric dental department on his second day of life for the management of a large intraoral mass that was disturbing breastfeeding (Fig 1A). The infant had been delivered by cesarian section with a weight of 3.54 kg, a height of 51 cm, and an Apgar score of 8 of 10. The hematologic and biochemical blood analysis results and the findings for all other systems were within the normal limits. The clinical examination revealed a

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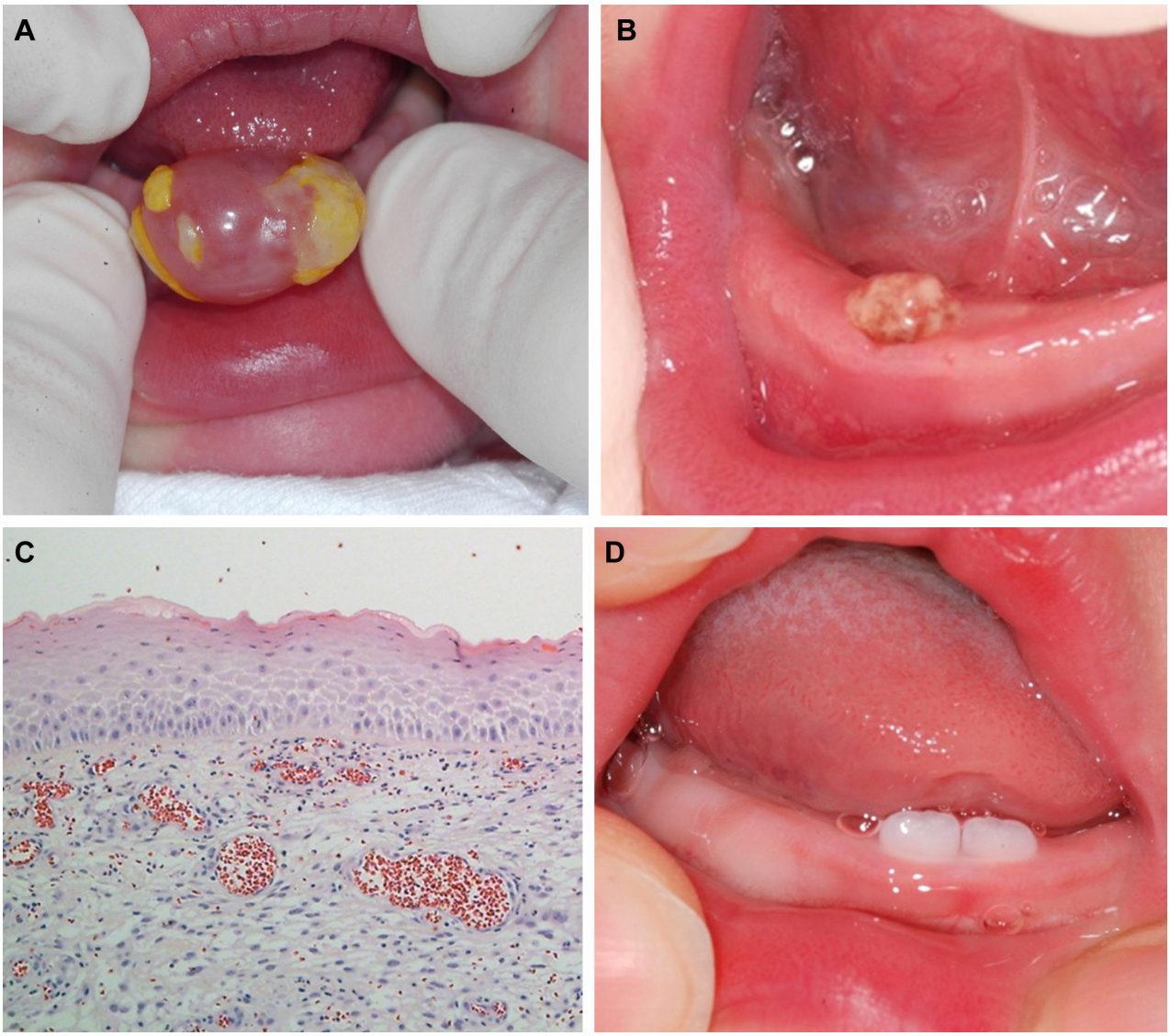

FIGURE 1. *A,* A male baby was referred for the management of a large intraoral mass on the lower anterior gum. The tumor was excised using Waterlase MD. *B,* The tissues were healing the day after surgery. The lesion was covered by a thin layer of connective tissue with an overlying keratinizing squamous epithelium. *C,* A delicate plexiform network of vessels was seen (hematoxylin and eosin stain, original magnification ×100). *D,* At the 3-month follow-up visit, healing was complete, no recurrence was seen, and no effects on the deciduous teeth were present.

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firm, lobulated, polypoid, and pedunculated tumor protruding from the lower anterior gingiva. Its dimensions were approximately 1.5 × 1.0 × 0.5 cm, and it was pink to red with a smooth surface. After the clinical evaluation, the differential diagnosis was CGCT, hematoma, and melanotic neuroectodermal tumor. Treatment consisted of surgical excision with the patient under local anesthesia using the Waterlase MD (Er, Cr:YSGG laser, BIOLASE, Irvine, CA) with a laser setting of 25 Hz and 1.5 W. Removal of the mass was performed by cutting the thin stalk, leaving some remnant tissues to avoid invasive surgery. Hemostasis was achieved immediately after removal, and regular breastfeeding began 3 hours after the surgery. The specimen was submitted to the Department of Pathology, Kyung Hee Medical Center, for histopathologic assessment.

Histopathologically, the tumor consisted of a proliferation of spindle cells with myxoid stroma and various size vascular channels. Mucosal ulceration and granulation tissue formation were also present. The overlying epithelium did not show pseudoepitheliomatous hyperplasia. However, typical atrophy of the rete ridges was observed (Fig 1C). The final histopathologic diagnosis confirmed the lesion was consistent with a CGCT.

On the day after surgery, the patient was breastfeeding normally (Fig 1B) and by his 3-month checkup, he