CASE REPORT OLGU SUNUMU

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Congenital Granular Cell Tumour of the Oral Cavity Causing Sucking and Feeding Difficulties in a Newborn

Yenidoğanda Emme ve Beslenme Güçlüğüne Neden Olan Oral Kavitenin Konjenital Granüler Hücreli Tümörü

Gaffari TUNÇ[a], Neşe KURT ÖZKAYA[b], Mahmut EKİCİ[c], Gözde KÖYLÜ[d], Hatice Reyhan EĞİLMEZ[e]

[a]Bursa Yüksek İhtisas Teaching Hospital, Clinic of Neonatology, Bursa, Türkiye
[b]Medicana Sivas Hospital, Clinic of Plastic Reconstructive and Aesthetic Surgery, Sivas, Türkiye
[c]Private Kolmed Hospital, Clinic of Pediatrics, Amasya, Türkiye
[d]Sivas Numune Hospital, Clinic of Pediatrics, Sivas, Türkiye
[e]Sivas Cumhuriyet University Faculty of Medicine, Department of Medical Pathology, Sivas, Türkiye

ABSTRACT Congenital granular cell tumors (CGCT) in newborns are rare benign tumors, growing on the alveolar ridge of the maxillary bone in newborns, mostly seen in females. Localization in the oral cavity can cause serious sucking problems resulting in nutritional difficulties and respiratory distress. Case is here presented of a female infant with a birth weight of 2,900 g born at 39 weeks gestation age with vaginal delivery. On physical examination, a lesion was observed 50x30x20 mm in size on the alveolar ridge of the maxillary bone protruding from the mouth. In the follow-up, infant experienced nutritional and breastfeeding problems but no respiratory distress. Tumor required surgery because it didn't regress spontaneously and was causing the nutritional deficiency. Early surgical intervention of such cases may prevent future sucking and feeding problems. In addition, CGCT seen in newborns is histopathologically different from adults, as it isn't immunoreactive for S-100 protein.

Keywords: Newborn; congenital granular cell tumor; sucking problems; S-100 immunostaining

ÖZET Yenidoğanlarda konjenital granüler hücreli tümörler [congenital granular cell tumors (CGCT)], yenidoğanlarda maksiller kemiğin alveolar sırtında büyüyen, çoğunlukla kızlarda görülen nadir benign tümörlerdir. Ağız boşluğundaki lokalizasyon, beslenme güçlüğü ve solunum sıkıntısıyla sonuçlanan ciddi emme sorunlarına neden olabilir. Burada, 39 haftalık gebelik haftasında vajinal doğumla dünyaya gelen 2.900 g doğum ağırlığında bir kız bebek sunulmuştur. Fizik muayenede, maksiller kemiğin alveolar sırtında ağızdan çıkıntı yapan 50x30x20 mm boyutlarında bir lezyon gözlenmiştir. İzlemde bebekte beslenme ve emzirme sorunları görülmüş ancak solunum sıkıntısı yaşanmamıştır. Tümör kendiliğinden gerilemediği ve beslenme yetersizliğine neden olduğu için ameliyat gerektirmiştir. Bu tür vakaların erken cerrahi müdahalesi ileride oluşabilecek emme ve beslenme sorunlarını önleyebilir. Ayrıca, yenidoğanlarda görülen CGCT, S-100 proteini için immünoreaktif olmadığı için histopatolojik olarak yetişkinlerden farklıdır.

Anahtar Kelimeler: Yenidoğan; konjenital granüler hücreli tümör; emme problemleri; S-100 immün boyama

The Greek term "epulis" means "on the gingiva" and as such, congenital granular cell tumor (CGCT) was first described as "congenital epulis" by Neumann in 1871.[1] A CGCT is a rare benign oral cavity tumor that presents at birth. Generally, females are affected 10 times more than males. It is seen in the maxilla 2-fold more than in the mandible. CGCT lesions can be of different sizes and can be pedunculated. The nature of this entity is not clear. CGCT is believed to be of neuronal origin.[2] It has no malignant potential and sometimes regresses spontaneously after birth.[3] CGCT in newborns is considered

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Correspondence: Gaffari TUNÇ
Bursa Yüksek İhtisas Teaching Hospital, Clinic of Neonatology, Bursa, Türkiye
E-mail: dr.gaffaritunc@gmail.com

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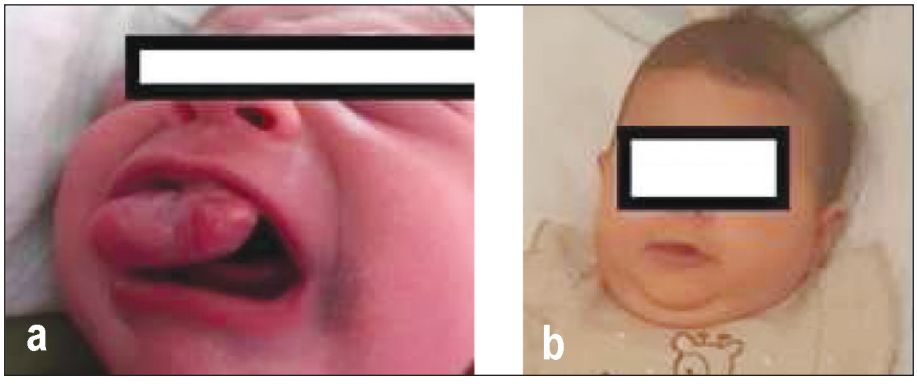

FIGURE 1: A: The appearance of the tumor in the oral cavity. The lesion is approximately 50x30x20 mm in the alveolar ridge of the maxillary bone in the newborn female. **B:** Postoperative appearance of the patient

to be a different entity from adults because of immunohistochemical features.[4]

CASE REPORT

In this case, a female infant was born at 39 weeks gestation age by vaginal delivery, with a birth weight of 2,900 g. There was nothing remarkable in the family history. The Apgar scores were 8 at 1 min and 9 at 5 min. On physical examination, there was observed to be a 50x30x20 mm lesion on the alveolar ridge of the maxilla bone (Figure 1A, B).

Laboratory results were normal. In the follow-up, the infant experienced nutritional and breastfeeding problems but no respiratory distress. Surgery was performed on postnatal day 12 by the plastic reconstructive and aesthetic surgery department. The lesion was excised and sent to a pathology laboratory. There were no postoperative complications. No recurrence was observed in the 6-month follow-up of the patient.

The macroscopic assessment revealed a smooth-surfaced polypoid lesion 50x30x20 mm in size, which was covered by gray-white mucosa. The histopathological findings were reported as a tumor mass of uniform appearance, consisting of large polyhedral histiocyte-like cells with small dark nuclei and abundant eosinophilic granular cytoplasms (Figure 2A, 2B). The lesion was covered by multilayered squamous epithelium on the surface; there was no atypia, necrosis, nor mitosis. No tumor was seen at the surgical margins. Tumor cells were found to be

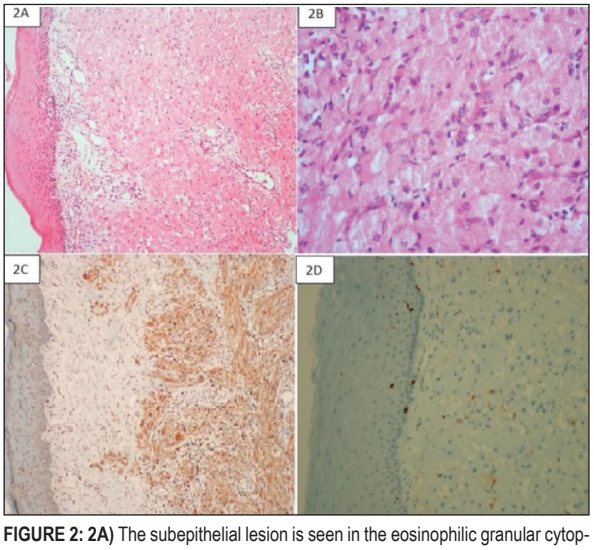

FIGURE 2: 2A) The subepithelial lesion is seen in the eosinophilic granular cytoplasm (H&E, x25); **2B)** High power field in figure 2A (H&E, x100); **2C)** There is immunoreactivity with CD68 (IHC, x25); **2D)** CGCT is S-100 protein negative (IHC, x25).

positive with the Periodik asit-Schiff stain. There was immunoreactivity with CD68 [immunohistochemical (IHCx25)] (Figure 2C). The lesion cells were not immunoreactive for S-100 protein, CD1a, and neuron specific enolas (IHC x 25) (Figure 2D). The parents gave verbal consent for publication of the case.

DISCUSSION

The clinical presentation of CGCT includes various sizes of lobular or oval, sessile, or pedunculated swelling covered with a flat, normal/reddish mucosal surface. Large lesions cause a mechanical obstruction to feeding and breathing in newborns.[5] The physical

examination and laboratory tests are normal.[6] The etiology of this tumor is not known. Generally, the mass is seen in an alveolar ridge of the maxillary bone in females. In the current case, a 50x30 mm lesion was determined on the alveolar ridge of the maxillary bone in a newborn female.

Differential diagnoses should be made with other congenital oral lesions. CGCT should be clinically differentiated from other congenital anomalies.[4] During pregnancy, oral cavity lesions can be identified in detail in utero examination. In delivery room management, oral cavity lesions should be handled cautiously. Early manipulation may be considered in the delivery room to provide airway patency and a complicated approach. Although cases are associated with polyhydramnios, neurofibromatosis, XXX chromosome, and polydactyly, CGCT is usually an isolated finding.[7]

The diagnosis must be confirmed by superficial ultrasound and computed tomography or magnetic resonance imaging.[8] In histological findings, CGCT is characterized by a proliferation of polygonal cells with eosinophilic, granular cytoplasm, and eccentric, bland appearing nuclei.[9] The immunohistochemical profile of the tumor in newborns is different from that of adults. There is no pseudoepitheliomatous hyperplasia. A positive reaction to carcinoembryonic antigens or Human Leukocyte Antigen (HLA)-DR antigens differentiates CGCT from other granular cell tumors. CGCT has been reported to be S-100-negative, and in the current case, the S-100 protein was negative.[9,10]

CGCT may rarely cause obstruction of the airway. The current case had no difficulty in respiration but the mass in the oral cavity was causing difficulty breastfeeding, with impaired sucking and feeding functions of the newborn. Therefore, oral feeding could be delayed. If CGCT causes breastfeeding and feeding problems, and there is respiratory distress, emergency surgical treatment is necessary.

Early surgical intervention in such cases may prevent future sucking and nutritional complications. Pathologically, a CGCT in newborns is considered different from the entity in adults, since it is not immunoreactive for the S-100 protein. Neonatologists need to consider CGCT in newborns with oral lesions and should aim for early diagnosis and referral for surgical intervention, then after surgery should help the mother to ensure adequate breastfeeding.

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Conflict of Interest

No conflicts of interest between the authors and / or family members of the scientific and medical committee members or members of the potential conflicts of interest, counseling, expertise, working conditions, share holding and similar situations in any firm.

Authorship Contributions

Idea/Concept: *Gaffari Tunç, Neşe Kurt Özkaya;* ***Design:*** *Gaffari Tunç, Hatice Reyhan Eğilmez;* ***Control/Supervision:*** *Hatice Reyhan Eğilmez, Gaffari Tunç, Gözde Köylü, Mahmut Ekici;* ***Data Collection and/or Processing:*** *Gaffari Tunç, Neşe Kurt Özkaya;* ***Analysis and/or Interpretation:*** *Gaffari Tunç, Neşe Kurt Özkaya, Gözde Köylü;* ***Literature Review:*** *Gaffari Tunç, Hatice Reyhan Eğilmez, Mahmut Ekici;* ***Writing the Article:*** *Gaffari Tunç, Hatice Reyhan Eğilmez;* ***Critical Review:*** *Gaffari Tunç, Hatice Reyhan Eğilmez;* ***References and Fundings:*** *Gaffari Tunç, Gözde Köylü, Hatice Reyhan Eğilmez.*

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