

Congenital granular cell lesion of the tongue: a report of two cases and review of the literature



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The congenital granular cell lesion most commonly occurs on the maxillary or mandibular alveolus of neonates. Extra-alveolar congenital granular cell lesion is exceptionally rare, with only 10 cases reported. Two additional cases occurring on the tongue are presented with a description of the clinical, histopathologic, and immunohistochemical features. The differential diagnosis is discussed, and the literature reviewed. (*Oral Surg Oral Med Oral Pathol Oral Radiol* 2016;122:e14-e18)

The congenital granular cell lesion (CGCL) is benign and most commonly presents on the maxillary or mandibular alveolus of neonates and is well documented in the literature.^{1,2} Occurrence on extra-alveolar sites is exceedingly rare with only 10 cases reported (nine lingual, one labial) (Table I).³⁻¹² The size of extra-alveolar lesions ranges from millimeters up to several centimeters and presents clinically as a lobular mass.³⁻¹² Large lesions cause feeding difficulties.^{3,7,11} Definitive treatment consists of excision, yet spontaneous resolution has been described.³⁻¹² There are no reports of recurrence or malignant transformation.³⁻¹² We present the clinical, histopathologic, and immunohistochemical findings of two cases of CGCLs, both occurring on the tongue of 2-day-old female infants. The literature regarding extra-alveolar lesions is reviewed.

CASE REPORTS

Case 1

The oral and maxillofacial surgery service at Long Island Jewish Medical Center was consulted by neonatal medicine to evaluate a large soft tissue lesion on the tongue of a 2-day-old female. She was born with no complications at full term by vaginal delivery. Her mother was in good health with no significant medical history and had had an uneventful pregnancy. The patient's mother reported interference with regular

feeding related to the lesion. On clinical examination, the child was in no apparent distress and was medically stable. Intraoral examination revealed a firm, nonpulsatile, pedunculated mass, measuring 2.0 × 1.5 cm, attached to the anterior ventral tongue (Figures 1 and 2). The mass appeared to prevent the infant from retracting her tongue into the oral cavity while at rest.

Excision of the mass was performed under general anesthesia with oral endotracheal intubation. Electrocautery was utilized to achieve hemostasis, and the wound was closed with 4-0 Vicryl simple interrupted sutures. The intact mass was placed in neutral buffered 10% formalin and submitted for histopathologic review.

Histopathologic review of the lesion showed an attenuated, parakeratinized, stratified squamous epithelium lacking rete ridges, with an underlying submucosal proliferation composed almost entirely of large, rounded, and polyhedral cells with small, dark, oval nuclei and abundant eosinophilic granular cytoplasm (Figure 3). The granular cells abutted the overlying epithelium. The stroma consisted of minimal fibrous tissue and rare vascular channels. Immunohistochemical evaluation for CD68 was positive (Figure 4), and S100 protein was negative (Figure 5). A diagnosis of congenital granular cell lesion was rendered.

There were no perioperative or postoperative complications. On postoperative day 1, the infant resumed breastfeeding without difficulty. At 1 week follow-up, the surgical site was healed, and the infant had no impairment of tongue mobility.

Case 2

A 2-day-old female infant presented to oral and maxillofacial surgery with a mass on the ventral tongue (Figure 6). The patient was asymptomatic and otherwise healthy. The pedunculated mass was smooth, mucosal colored, soft to palpation, and measured 0.8 × 0.6 × 0.4 cm. The lesion was excised under local anesthesia without complication and submitted for histopathologic evaluation.

Microscopic evaluation revealed an attenuated epithelium lacking rete ridges, overlying a sheet-like proliferation of granular cells possessing small hyperchromatic nuclei and abundant eosinophilic cytoplasm. Immunohistochemical studies showed positivity for vimentin and CD68. Antibodies directed against S100 and α -smooth muscle actin were negative. The lesion was diagnosed as congenital granular cell lesion. At 8 months' follow-up, the patient was free of disease with no signs of recurrence.

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Received for publication Jun 24, 2015; returned for revision Sep 29, 2015; accepted for publication Oct 9, 2015.

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2212-4403/\$ - see front matter

<http://dx.doi.org/10.1016/j.oooo.2015.10.013>

Table I. Cases of extra-alveolar congenital granular cell lesion

Case	Year	Author	Gender	Location	Concurrent lesion	Size (cm)
1	1957	Atterbury	Male	Tongue	No	2.0
2	1975	Cussen	Female	Tongue	No	1.3
3	1975	Dixter	Female	Tongue	Yes	2.0
4	1987	Ophir	Female	Tongue	Yes	1.0
5	1997	Loyola	Female	Tongue	Yes	3.0
6	2001	Yavuzer	Female	Tongue	Yes	1.0
7	2007	Senoo	Female	Tongue	No	0.4
8	2011	Kayiran	Female	Tongue	No	2.0
9	2011	Childers	Unspecified	Lip	No	Unspecified
10	2014	He	Female	Tongue	No	2.0
11	2015	Current case 1	Female	Tongue	No	2.0
12	2015	Current case 2	Female	Tongue	No	0.8



Fig. 1. Clinical image of case 1 depicting a multilobular mass emanating from the oral cavity.



Fig. 2. Clinical image of case 1 demonstrating location of mass on the anterior tongue (same patient as in Figure 1).

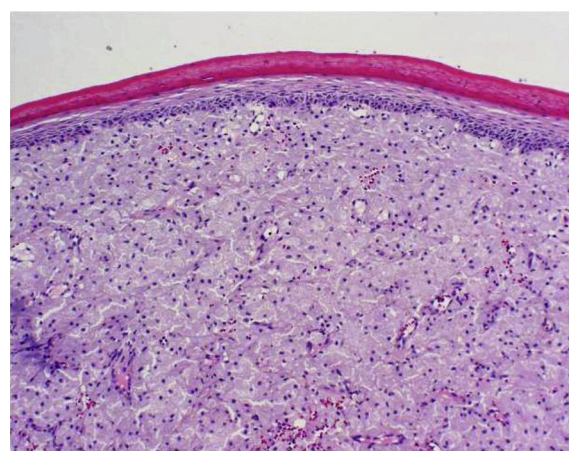


Fig. 3. Excision specimen of mass from case 1 demonstrates an attenuated, parakeratinized, stratified squamous surface epithelium. Note the absence of rete ridges. The submucosa shows a proliferation of large, rounded, and polyhedral cells with small, dark nuclei and abundant eosinophilic granular cytoplasm with a vascular stroma (hematoxylin and eosin $\times 40$ original magnification). A high-resolution version of this slide for use with the Virtual Microscope is available as eSlide: VM01477.

DISCUSSION

The CGCL most commonly occurs on the maxillary alveolar process of female infants.^{1,2} It can be confused with a granular cell tumor (GCT), which has some

overlapping histopathologic features but is a separate and distinct clinicopathologic entity.^{1,2,7} The GCT occurs in adults, most commonly on the tongue as a submucosal nodule.^{2,12} Microscopic evaluation of the GCT demonstrates a thickened surface epithelium that may show pseudoepitheliomatous hyperplasia (PEH).^{2,12} This is in contrast to the CGCL, which possesses an attenuated surface epithelium, lacking rete ridges.²⁻¹² Like the CGCL, the GCT demonstrates an underlying population of granular cells. Evaluation of the granular cells for S100 protein by immunohistochemistry is positive in the GCT but negative in the CGCL.^{1,2,13,14} The GCT is also treated by conservative excision.²

Alveolar CGCL is well documented in the literature.^{1,2} In contrast, only 10 cases of extra-alveolar CGCLs have been reported.³⁻¹² Our review of the

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