A 27-month-old otherwise healthy African American girl presented to the pediatric otolaryngology clinic for second opinion of a persistently disfiguring left buccal mass. At 3 weeks of age, she developed asymptomatic left-sided facial swelling which gradually enlarged, resulting in progressive asymmetry of the face. She underwent formal evaluation of the swelling at 5 months of age initially at an outside institution. Review of magnetic resonance imaging (MRI) demonstrated a large T1 hypointense, T2 hyperintense, unilocular, cystic lesion located in the left buccal region. Certain aspects of the peripheral rim appeared brighter than others in postcontrast scan. Such absence of homogenous circumferential rim enhancement suggested that hyperintense regions were consistent with adjacent vessels. As such, the lesion was presumed to be a macrocystic lymphatic malformation (Figure 1). The patient was treated with 2 rounds of doxycycline sclerotherapy without resulting change in size of the lesion, according to repeat MRI (Figure 2).

At our institution, careful intraoral and extraoral inspection by a pediatric otolaryngologist revealed a 4.5 cm × 3.5 cm × 1.5 cm swelling adjacent to the left buccal and left retromaxillary fat pads. On palpation, the lesion was supple, nontender, and freely movable without involvement of the overlying skin or intraoral mucosa. Excisional biopsy of the lesion was planned to confirm the diagnosis with surgical resection for definitive treatment. Following subtotal removal, the lesion was sent for histopathological analysis, revealing a cystic cavity lined by stratified squamous epithelium, devoid of cutaneous adnexa, and confirmed squamous pearls (1-2 mm) present in the capsule wall. The central lumen contained friable, yellow-brown contents consisting of degenerated keratin. There was no evidence of vascular proliferation. This pathology, in correlation with clinical findings, supported a final diagnosis of epidermoid cyst. At the first postoperative visit (1 month after surgery), the patient had mild ipsilateral facial weakness, which improved by 3 months postoperatively. Subsequent follow-up 1 year postoperatively demonstrated mild persistent weakness of the lower division of the face at rest and no proliferation of the residual portion of the lesion.

Dermoid and epidermoid cysts are rare developmental cysts known to occur throughout the body, with 7% of cases encountered in the head and neck.1 The most common intraoral site is the floor of the mouth.2 Occasionally, lesions have been reported to crop up in the tongue, lips, nose, and intraorally in the maxilla and mandible.5,6 Epidermoid cysts of the buccal space are exceedingly rare. Only 6 cases have been reported in the literature, and all are among adult patients ranging from 25 to 65 years of age.6,12 Lesions arising in the buccal mucosa commonly present as a progressively enlarging, soft, mobile growth without a history of local trauma. Size at presentation varies considerably (15-45 mm in greatest dimensions), with the cyst showing predilection for the left buccal space. Generally, disease goes unnoticed until sizable mass effect produces some degree of cosmetic deformity or functional limitation due to displacement of intraoral structures.4

Clinically and radiologically, epidermoid cysts may mimic several other pathologies in the buccal space, including odontogenic abscesses of the buccal or masseteric space; developmental cysts; lipomas, lymphangiomas, hemangiomas, or pleomorphic adenomas; mucoceles; lymphatic malformations; or less commonly, malignant tumors (of epidermal or mesodermal origin).

To improve diagnostic accuracy, evaluation using contrast-enhanced MRI or ultrasonography is recommended to delineate epidermoid cysts from lesions with prominent vascularization or other defining characteristics. On axial-cut contrast-enhanced MRI, epidermoid cysts appear as a well-
circumscribed mass with low signal intensity in T1-weighted images and high signal intensity in T2-weighted images, similar to many other lesions. Ultrasonography reveals a well-demarcated capsule with a heterogeneous interior due to the presence of keratinized fluid. Thorough review of all imaging planes may be indicated for preoperative planning to determine the extent of the lesion and to facilitate visualization of its relationship to surrounding structures.

A definitive diagnosis is achieved by histopathologic analysis. As in this case, the patient’s prior sclerotherapy treatment had complicated interpretation of her posttreatment MRI, and thus, histopathology was required to obtain an accurate preliminary diagnosis. Initial assessment of pathology is important to determine appropriate treatment modality, which is likely adapted to each clinical case. If an epidermoid cyst is suspected, incisional or aspiration biopsy may be useful to guide method of intervention.

In general, epidermoid cysts do not undergo malignant transformation. For lesions that are smaller or asymptomatic, watchful waiting may be considered. Total resection usually results in resolution of symptoms without anticipated recurrence.

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References

1. New GB. Dermoid cysts of the head and neck. Surg Gynecol Obstet. 1937;65(2):48-55.
2. Sabhalok SS, Shetty LS, Sarve PH, Setiya SV, Bharadwaj SR. Epidermoid and dermoid cysts of the head and neck region. Plast Aesthet Res. 2016;3(suppl 1):347-350.
3. Worley CM, Laskin DM. Coincidental sublingual and submental epidermoid cysts. J Oral Maxillofac Surg. 1993;51(7):787-790.
4. Shear M, Speight P. Cysts of the Oral and Maxillofacial Regions. 4th ed. Hoboken, NJ: John Wiley & Sons; 2008.
5. Flom GS, Donovan TJ, Landgraf JR. Congenital dermoid cyst of the anterior tongue. Otolaryngol Head Neck Surg. 1989;100(6):602-605. doi:10.1177/019459988910000614.
6. Schneider L, Mesa ML. Epidermoid cysts of the buccal mucosa. Quart Nat Dent Associat Inc. 1978;36(2):39.
7. Gutmann J, Cifuentes C, Gandulfo P, Guesalaga F. Intradermal nevus associated with epidermoid cyst in the mucous membrane of the cheek. Oral Surg Oral Med Oral Pathol. 1978;45(1):76-82. doi:10.1016/0030-4220(78)90226-8.
8. Rajayogeswaran V, Eveson J. Epidermoid cyst of the buccal mucosa. Oral Surg Oral Med Oral Pathol Oral Radiol. 1989;67(2):181-184. doi:10.1016/0030-4220(89)90326-5.
9. Ozan F, Polat HB, Ay S, Goze F. Epidermoid cyst of the buccal mucosa: a case report. J Contemp Dent Pract. 2007;8(3):90-96.
10. Kini YK, Kharkar VR, Rudagi B, Kalburge JV. An unusual occurrence of epidermoid cyst in the buccal mucosa: a case report with review of literature. J Oral Maxillofac Surg. 2013;12(1):90-93. doi:10.1007/s12663-011-0188-y.

11. Costa FWG, Carvalho FSR, Chaves FN. Epidermoid cyst arising in the buccal mucosa: case report and literature review. Acta stomatologica Croatica. 2015;49(1):65-73. doi:10.15644/asc49/1/9.
12. Ichikawa D, Ohba S, Yoshimura H, Matsuata S, Imamura Y, Sano K. Epidermoid cyst of the buccal mucosa diagnosed by magnetic resonance imaging and ultrasonography: a case report and review of the literature. Oral Health Dent Managl. 2015;14(4):0-4.
13. Curtin HD, Som PM. Head and Neck Imaging. 5th ed. New York, NY: Mosby; 2011;1058.
14. Kim HC, Han MH, Moon MH, Kim JH, Kim IO, Chang KH. CT and MR imaging of the buccal space: normal anatomy and abnormalities. Korean J Radiol. 2005;6(1):22-30. doi:10.3348/kjr.2005.6.1.22.
15. Reddy A, Kreicher KL, Patel NA, Schantz S, Shinhar S. Pediatric epidermoid cysts masquerading as ranulas: a case series. Int J Pediatr Otorhinolaryngol. 2016;81:26-28. doi:10.1016/j. ijporl.2015.11.031.
16. Jamal N, Ahmed S, Miller T, et al. Doxycycline sclerotherapy for pediatric head and neck macrocystic lymphatic malformations: a case series and review of the literature. Int J Pediatr Otorhinolaryngol. 2012;76(8):1127-1131. doi:10.1016/j.ijporl.2012.04.015.
17. Wiegand S, Eivazi B, Zimmermann AP, Sesterhenn AM, Werner JA. Sclerotherapy of lymphangiomas of the head and neck. Head Neck. 2011;33(11):1649-1655. doi:10.1002/hed.21552.
18. Cameron DS, Hilsinger RL. Squamous cell carcinoma in an epidermal inclusion cyst: case report. Otolaryngol Head Neck Surg. 2003;129(1):141-143. doi:10.1016/S0194-59980300466-2.