BRIEF ARTICLES

Eruptive Oral Mucoceles in a Neonate

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ABSTRACT

Mucoceles are one of the most common benign mucosal growths in the oral cavity. They may be under-recognized in children. Mucoceles infrequently present in groups. We present an unusual case of eruptive superficial mucoceles in a 6-day old male that served as a diagnostic dilemma.

CASE PRESENTATION

A 6-day-old male presented to the Emergency Department (ED) with a 1-day history of multiple blisters on the lower lip noticed by the mother while breastfeeding. The patient's medical history was notable for spontaneous vaginal delivery at term to a G1P1 mother with history of labial herpes resolved without treatment. The mother reported taking only multivitamins, while there was no medication history for the baby. The patient was afebrile and hemodynamically stable on arrival to the ED, and parents reported normal feeding, bowel movements, and sleep. No rash in other parts of the body or trauma was reported.

Physical examination revealed the presence of multiple 2-4 mm clustered vesicles on the lower lip mucosa filled with yellow to clear fluid (Figure 1). Viral culture, bacterial culture, cerebrospinal fluid culture (CSF), Herpes Simplex Virus types 1,2 Polymerase Chain Reaction (PCR), and Direct Fluorescent Antibodies (DFA) were obtained, and the patient was started on empiric IV acyclovir due to high suspicion of oral herpes simplex. Dermatology was consulted at this point.

The patient was admitted to the pediatric floor for close monitoring on IV acyclovir. DFA, viral cultures, and CSF cultures were subsequently negative. On day 6 of admission, the patient developed a new solitary vesicle on the lower lip (Figure 2), and repeat HSV PCR was performed. On day 9 of admission, the HSV PCR was negative, and 2 new vesicles appeared. Otolaryngology was consulted for a lip biopsy (Figure 3 A & B) given continued concern for possible herpes despite normal studies.

Histopathology demonstrated a superficial, unlined dermal cyst with mucin (Figure 3A), highlighted by colloidal iron stain (Figure 3B), suggestive of a superficial variant of a mucocele. The patient was discharged

without further treatment and did not develop any subsequent oral lesions.

Figure 1. Clinical photograph of lower labial mucosa showing multiple, grouped vesicles

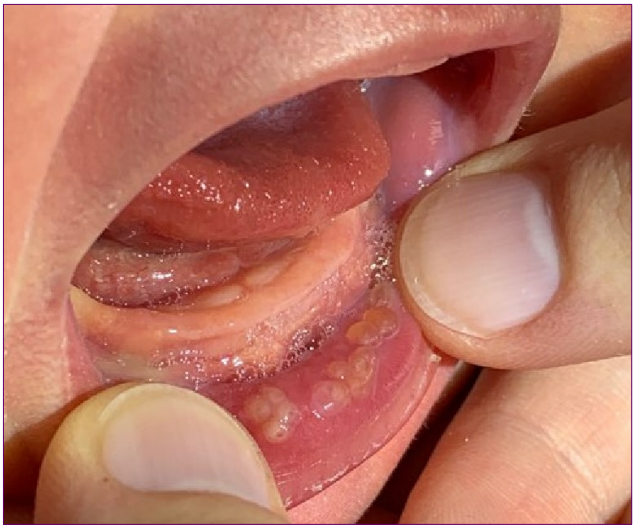
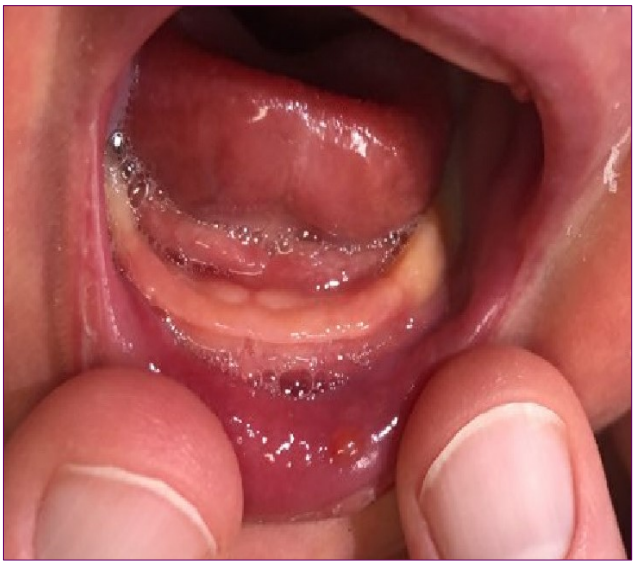


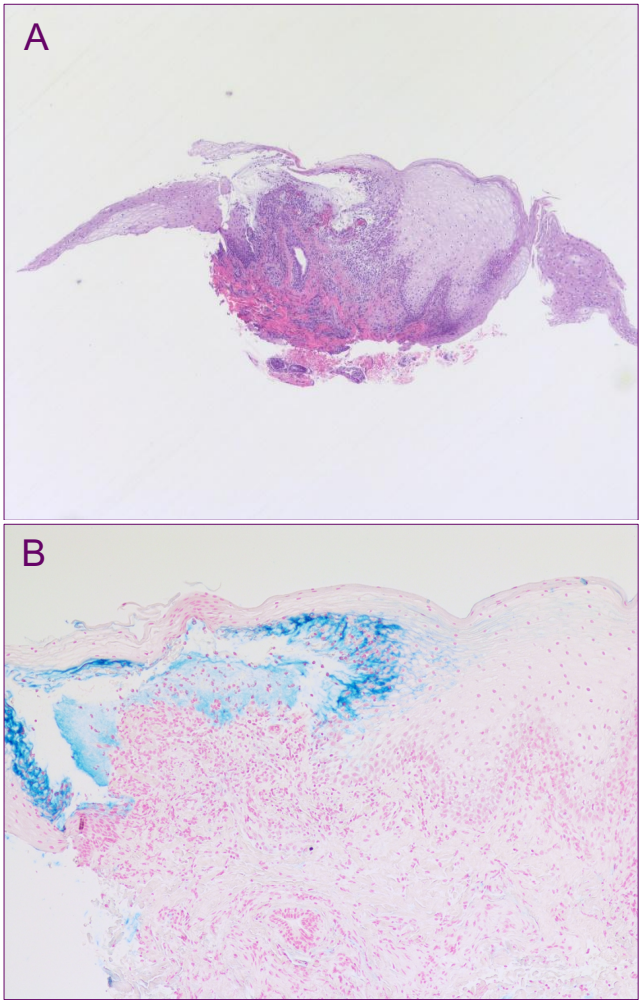
Figure 2. Solitary vesicle on lower lip containing thin yellow fluid on the lower labial mucosa



DISCUSSION

Mucoceles are common, benign mucosal lesions. They are often caused by trauma with rupture of the salivary ducts and extravasation of mucus into the surrounding

Figure 3. (A) Hematoxylin & eosin stain (4x) of the biopsy demonstrates reveals stratified squamous epithelium with underlying connective tissue consisting of large cyst in the lamina propria with mucinous material splitting apart collagen bundles. **(B)** Colloidal iron stain (10x) highlights mucin within superficial cyst.



lamina propria, though they can also possess an epithelial lining .[1-5] Although patient of any age can be affected, the peak frequency occurs in the 1st to 3rd decade of life.[6-8] The prevalence in children has been reported as low as 0.08%, however another large cohort reported by Shapira et al. (2014), demonstrated 7.1% occurring under one year of age.[7,9] A subset of mucoceles, as initially described by Eveson (1988), has been classified as “superficial,” which defines them as small, tense, translucent vesicles on the oral mucosa.[10] Superficial mucoceles usually occur in the soft palate, retromolar pad, or posterior

buccal mucosa and have been identified as the most common variant.[7,11]

There are two mechanisms for developing mucoceles: extravasation and retention. The prevailing extravasation type, which presents frequently in children, is due to the rupture of a minor salivary gland duct and leakage of mucin into the adjacent tissue which then becomes encapsulated by granulation tissue. Retention-type mucoceles develop due to an obstruction of the salivary duct with either heavy mucin or small sialolith.[13,14] In neonatal mucoceles, trauma to a salivary duct may be attributed to sucking fingers in utero, passage through the delivery canal, forceps use during delivery in assisted vaginal delivery, or manipulation of the neonate by nurses after birth.[9] There is no evidence that breast or bottle feeding contributes to the risk of mucocele development, and most cases report solitary mucoceles. There are few cases of multiple mucoceles.[11,15]

Though there do not appear to be established guidelines for the management for mucoceles, a review of the literature suggests most authors prefer conventional surgical excision with the removal of the adjacent salivary glands for definitive removal and preventing relapse. Other treatment modalities have been tried with varying success include cryosurgery, intra-lesion corticosteroid injection, micro-marsupialization, and CO2 laser.[12] In our case, no treatment was provided as lesions ruptured spontaneously with no recurrence on follow up.

CONCLUSION

Multiple eruptive superficial mucoceles can mimic vesiculobullous lesions and should be considered in the differential diagnosis of oral blisters in a neonate once infectious and immunobullous causes have been eliminated.

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