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CASE REPORT

Buccal salivary cysts in association with a congenital parotid salivary fistula

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CT sialography

Summary Congenital parotid salivary fistulas are unusual entities that can arise from accessory parotid glands or, even more infrequently, from normal parotid glands via an aberrant Stensen's duct. A unique case of a congenital parotid salivary fistula presented in a four-week-old infant as polycystic swelling of the buccal region, with cyst contents draining to a cutaneous pit near the oral commissure. This patient offered an initial diagnostic challenge until it became evident that the drainage represented a salivary fistula. The case is used here to highlight the overlapping clinical and pathologic features shared between type I branchial cleft anomalies and congenital salivary cysts. This case is the second report of buccal salivary cysts in structural association with a congenital parotid salivary fistula and the first such case in which a normal Stensen's duct orifice has been preserved.

1. Introduction

The differential for benign congenital cysts of the parotid region includes type I branchial cleft cysts, lymphatic malformations, salivary cysts, and dermoid cysts. The association of a connected draining cutaneous pit suggests a diagnosis of a type I branchial cleft anomaly when such lesions are encountered. Congenital salivary fistulas draining to the skin are more unusual entities than type I branchial cleft anomalies and are often not associated with cysts. They present as cutaneous pits arising from the parotid gland, submandibular gland, or ectopic salivary tissue [1], with labial pits usually associated with minor salivary glands. Occasional salivary fistulas can arise from accessory parotid glands, which are themselves common entities, present in 21% of anatomic specimens [2]. Normally these accessory glands merely drain to the Stensen's duct via two or three direct connections. However, multiple case reports exist of accessory parotid glands giving rise to congenital

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salivary fistulas to the cheek skin and various other sites [3—5].

Congenital salivary fistulas arising directly from the parotid gland are exceedingly rare, and their structural association with congenital salivary cysts outside the parotid gland is nearly unprecedented in the literature. Here we present a unique case of a congenital parotid salivary fistula presenting with pronounced polycystic swelling of the cheek and draining to a cutaneous pit near the oral commissure. The diagnostic evaluation, treatment planning, and histologic interpretation of this lesion all posed challenges that are described here. The case is reviewed in context of the scant existing literature on congenital salivary cysts and fistulas.

2. Case report

A four-week-old healthy term female infant with unremarkable prenatal history presented to our emergency department one day following acute onset of large right cheek swelling in association with an exam finding of a cutaneous pit just lateral to the right oral commissure (Fig. 1). There were no signs of infection noted, and an ultrasound demonstrated three submucosal cystic structures within the left buccal region. The patient was managed conservatively, with followup planned pending further imaging. However, two days later, the patient was admitted with purulent drainage from the right cheek pit, from which cultures grew *P. mirabilis* and community-acquired oxacillin-resistant *S. aureus*.

Treatment with intravenous antibiotics and expression of purulence from the cyst produced rapid resolution of infection, although residual buccal swelling anterior to the Stensen's duct orifice persisted. Magnetic resonance imaging (MRI) was performed, revealing three buccal cysts located anterior to the ramus of the mandible at a submucosal location. The favored diagnoses at this point included an unusually placed first branchial cleft anomaly versus a lymphatic malformation with an atypical drainage tract.

Despite resolution of infection, these cysts continued daily to accumulate fluid, which could be expressed as a turbid liquid resembling saliva. An amylase level was thus sent on the fluid and was assayed at 1300 IU/L. This finding prompted a computed tomography (CT)-sialogram to define the structural relationships between the cysts, the pit, the native Stensen's duct, and the parotid gland. Cannulation and injection of either the native Stensen's duct or the cheek skin pit directly filled the same three buccal submucosal cysts with contrast material (Fig. 2). Further injection of the

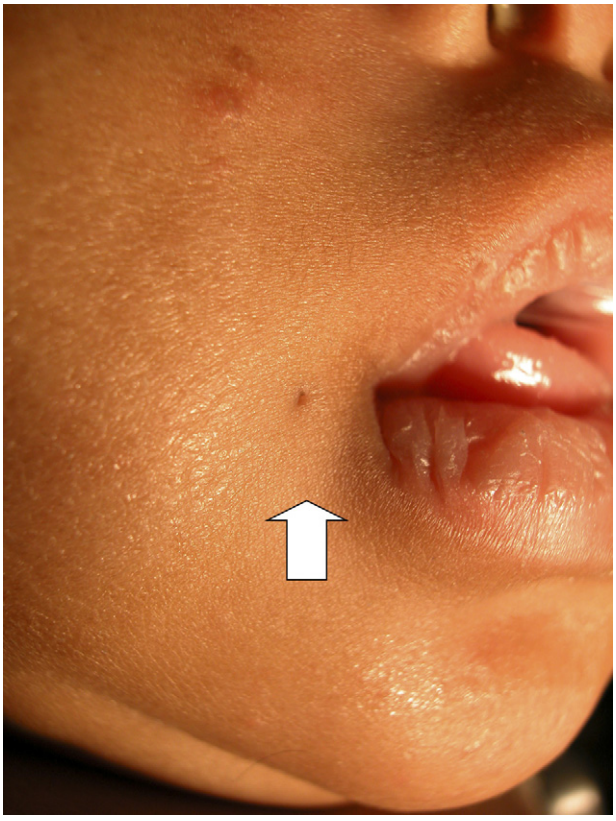

Fig. 1 Location and appearance of cutaneous pit just lateral to oral commissure (arrow).

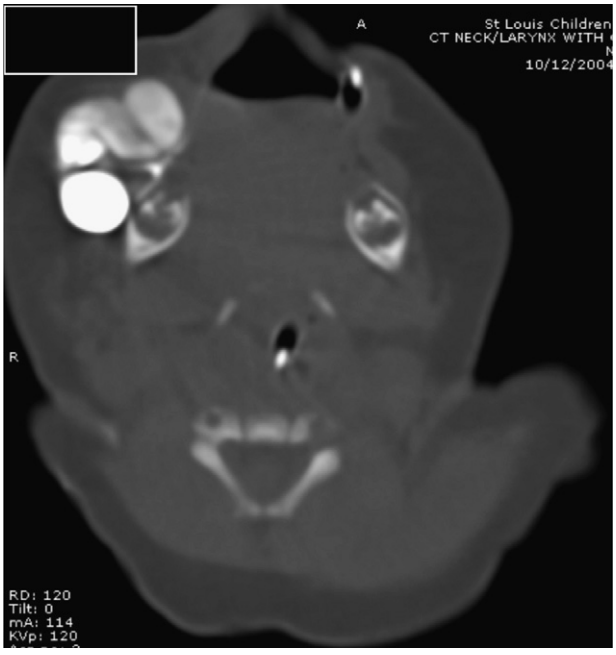

Fig. 2 CT sialogram showing contrast injection of either the oral commissure pit or the Stensen's duct papilla fills three buccal submucosal cysts.