An Unusual Presentation of a Lingual Dermoid Cyst

Megan L. Durr, MD\textsuperscript{1}; Annemieke van Zante, MD, PhD\textsuperscript{2}; Steven D. Pletcher, MD\textsuperscript{1}

\textsuperscript{1}Department of Otolaryngology – Head & Neck Surgery
\textsuperscript{2}Department of Pathology
University of California - San Francisco

ABSTRACT

INTRODUCTION

Dermoid cysts are rare benign lesions that can be found throughout the body. Approximately 7% of dermoid cysts occur in the head and neck and 1.6% occur in the oral cavity.\textsuperscript{1} The floor of the mouth is the most common site involved in the mouth, but dermoid cysts have been reported in the tongue, lips, and buccal mucosa.\textsuperscript{2} Histologically, Meyer divided cysts of the floor of mouth and tongue into three groups: epidermoid, dermoid, and teratoid.\textsuperscript{1} All three of these lesions have a cystic component lined by stratified squamous epithelium.

Dermoid cysts can be differentiated from epidermoid cysts by the additional finding of a variable number of skin appendages, including hair follicles and sebaceous glands. Dermoid cysts are frequently filled with desquamated epithelial cells, keratin debris, and hair.\textsuperscript{4} Teratoid cysts (cystic teratomas) are similarly composed of squamous lined cysts with skin appendages with the additional finding of mesodermal and endodermal elements such as muscle, bone, respiratory and gastrointestinal tissues.\textsuperscript{3,5,6} Although dermoid cysts represent a histologically distinct entity, the term “dermoid” is typically used to loosely indicate all three categories.

RESULTS:

The patient is a 48 year old man with a twenty year history of recurrent tongue and floor of mouth abscesses, requiring a tracheotomy and multiple hospitalizations for drainage. An MRI following his most recent infection revealed a 1.8 cm lesion within the tongue musculature. He underwent surgical resection of this lesion. Pathology showed a cyst lined by benign squamous mucosa with adjacent sebaceous glands, consistent with a dermoid cyst. The patient had no recurrent abscesses two years after surgical excision of the cyst.

CONCLUSIONS:

Dermoid cysts are rare lesions of the head and neck that usually present as asymptomatic, slow-growing masses. This case demonstrates that dermoid cysts may present later in life with recurrent acute infections.

CASE PRESENTATION

We present the case of a 48 year old man with a lingual dermoid cyst presenting as recurrent Ludwig’s Angina requiring tracheotomy. The patient had a twenty year history of recurrent tongue and floor of mouth abscesses. The first episode occurred at age 26, when he presented to an outside hospital with three days of pain and progressive floor of mouth and submental swelling associated with hoarseness of breath. He underwent an urgent tracheotomy with incision and drainage of a floor of mouth abscess. He was subsequently decannulated and remained asymptomatic for 10 years.

He had two similar episodes of relatively mild floor of mouth swelling 10 and 15 years after the initial episode. These episodes subsided after a course of oral antibiotics. In February of 2008, he presented to our institution with pain and floor of mouth swelling. His CT scan during this episode showed a rim-enhancing fluid collection within the tongue musculature with extension into the floor of the mouth (Figure 1). He underwent transoral incision and drainage and his symptoms resolved after two weeks of antibiotic treatment.

After resolution of symptoms, the patient had no palpable masses or lesions in the tongue. An MRI scan performed two months following resolution of symptoms demonstrated a 1.8 cm lesion within the genioglossus muscle that had low T1 signal and intermediate T2 signal, consistent with a dermoid cyst (Figure 2).

The lesion was excised using a midline sagittal glossotomy while maintaining the integrity of the dorsal tongue to minimize the risk of postoperative articulation deficits. The lesion had a small sinus tract which extended to the surface of the dorsal tongue and this was removed along with the cyst. He had no articulation deficits on follow-up examination, and has had no recurrent symptoms two years after surgical excision of the cyst.

Gross pathological examination revealed a 2.3 cm x 1.7 cm x 0.7 cm cyst filled with keratin debris and surrounded by a fibrous scar tissue. Macroscopic examination of the specimen revealed a benign squamous epithelial lining with adjacent sebaceous glands consistent with a dermoid cyst (Figures 3 and 4).

DISCUSSION

In a review of 1495 cases, Erich and Johnsen found that the majority of dermoid cysts are found in the sacral region and include pilonidal cysts (44.4%). Forty-two percent of dermoid cysts are found in the ovaries, with only 7% in the head and neck.\textsuperscript{1} In the head and neck, dermoid cysts are most common in the periorbital region (50%), followed by the floor of mouth, submental and submandibular regions (23%), frontal and neck regions (14%), and nasal regions (13%).\textsuperscript{1} Dermoid cysts of the body of the tongue are rare, and only twenty one cases have been reported in literature, all in pediatric patients.\textsuperscript{3,5,7} To our knowledge, this is the first case report of a lingual dermoid cyst in an adult. This is also the first reported case of a lingual dermoid cyst presenting as severe recurrent Ludwig’s angina requiring tracheotomy.

The broad differential diagnosis of sublingual/lingual lesions includes: ranula, lymphatic malformation, dermoid cyst, epidermoid cyst, neurofibroma, hemangioma, lingual thyroid, heterotopic gastrointestinal cyst, duplication foregut cyst and infection.\textsuperscript{5} Given this differential diagnosis, bimanual palpation and conventional radiography are not always sufficient in making the correct diagnosis.

Computed tomography and magnetic resonance imaging allow for precise localization of the lesion in relationship to surrounding musculature, and enable the surgeon to choose the most appropriate surgical approach, especially for very large lesions.\textsuperscript{2} Dermoid cysts are iso- or hypointense to muscle on T1-weighted images and hyperintense or heterogeneous on T2-weighted images.\textsuperscript{8}

Surgical excision is the treatment of choice for lingual dermoid cysts. Lingual dermoid cysts are generally easily accessible transorally. The midline sagittal glossotomy is commonly used for lingual dermoid cysts and tumors in the posterior one-third of the tongue. This approach minimizes the risk of damage to the submandibular ducts and decreases blood loss as it divides the tongue along the midline fibrous septum avoiding the more laterally located lingual vessels.\textsuperscript{8}

The prognosis of dermoid cysts is excellent, and recurrence is unlikely with complete excision.\textsuperscript{5} Malignant changes in dermoid and teratoid lesions are rare, but have been reported in a sublingual dermoid cyst and a teratoid cyst of the floor of mouth.\textsuperscript{8}

CONCLUSIONS

Dermoid cysts are rare lesions of the head and neck that usually present as asymptomatic, slow-growing masses in the pediatric population. Surgical excision is the treatment of choice for these lesions. We report the first case of a lingual dermoid cyst in an adult who presented with recurrent infection of the tongue and floor of mouth.

REFERENCES