High Grade Dysplasia of an Enteric Duplication Cyst of the Anterior Tongue

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ABSTRACT

Enteric duplication cysts are heterotopic rests of gastrointestinal mucosa. While these lesions have been found to occur along the entire digestive tract, involvement of oral cavity is excessively rare. The anterior tongue is the most commonly reported site for this subset and typically presents early in life secondary to mass effect leading to difficulties with swallowing, speech, or respiratory compromise. This case demonstrates not only a delayed presentation of symptoms but also raises the questions regarding the optimal management of these lesions in the face of significant dysplasia.

DISCUSSION – CONT’D

Histologically, these cysts are lined by gastric or intestinal epithelium that can be found in conjunction with squamous or respiratory epithelium1. While the vast majority of these oral cavity lesions are reported to be benign, there has been a report of adenocarcinoma arising in a duplication cyst in the floor of the mouth1. Due to the rarity of this disease, optimal management is not known. While complete excision is recommended by most, this may not be feasible in large lesions, as it was in this case, without causing excessive morbidity. The finding of high-grade dysplasia further complicates decision making in this patient. While pathologic analysis has the potential to be skewed in the face of active inflammation, there is a very real potential for malignant change. Ultimately, the decision for observation was left to the patient after a discussion of treatment approaches and the possible morbidity associated with each were explained. He has demonstrated no evidence of malignant disease on PET/CT imaging or clinical exam after 8 months, but has subsequently been lost to follow up.

CONCLUSIONS

Enteric duplication cysts affecting the oral cavity are rare lesions. While typically presenting in childhood, some may not become symptomatic until later in life. Optimal management is still being elucidated as experience grows with this clinical entity. Complete surgical excision is recommended when undue morbidity is not expected.

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REFERENCES