

Evaluation and management of benign, non-congenital tongue masses in children[☆]

Nina Lisbeth Shapiro^{a,*}, David J. Malis^b, Douglas F. Brewster^c,
Donald B. Kearns^d

^a *Pediatric Otolaryngology, Division of Head and Neck Surgery, UCLA School of Medicine, 10833 LeConte Avenue, 62-158 CHS, Los Angeles, CA 90095-1624, USA*

^b *Otolaryngology–Head and Neck Surgery Service, Brooke Army Medical Center, San Antonio, TX 78234-6200, USA*

^c *Department of Otolaryngology–Head and Neck Surgery, Naval Medical Center, San Diego, CA 92134-5000, USA*

^d *Pediatric Otolaryngology, Suite 402, Children’s Hospital, 3030 Children’s Way, San Diego, CA 92123-4228, USA*

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Abstract

Lingual tumors are rare, primarily benign, lesions in the pediatric population. Congenital lesions, such as hemangiomas, lymphatic malformations, dermoids, hamartomas and thyroglossal ducts cysts, are seen more commonly. Primary, non-congenital lingual neoplasms are less common in children. We present three patients with benign lingual neoplasms. Evaluation, management, pathology and follow-up are discussed. © 1999 Elsevier Science Ireland Ltd. All rights reserved.

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1. Introduction

Tongue masses in children are rarely encountered. When seen, they are most commonly con-

genital in origin. Congenital lesions in the anterior region may be hemangiomas or lymphatic malformations. Posterior and midline congenital lesions include thyroglossal duct cysts, lingual thyroid gland, dermoids, choris-tomas, and heterotopic gastric cysts. The location of these lesions in the region of the foramen cecum is secondary to embryologic disruption at the junction of the anterior two-thirds and posterior one-third of the tongue during development [1].

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* Corresponding author. Tel.: +1-310-825-2749; fax: +1-310-206-6688.

E-mail address: nshapiro@ucla.edu (N.L. Shapiro)

Benign tongue lesions that are not secondary to embryologic disturbance are quite rare. We describe three children who presented with benign, non-congenital lingual neoplasms. Diagnostic evaluation included radiologic imaging to evaluate extent of the mass and to rule out concomitant thyroid abnormalities. Complete excision is most often curative. Close follow-up is indicated to monitor for postoperative recurrence.

2. Case 1

A five-year-old male presented with a tongue base mass noted since early infancy. It did not interfere with his airway, oral intake, or speech. Examination revealed a 1.0-cm well-mucosalized pedunculated mass, just lateral to the region of the foramen cecum (Fig. 1). Thyroid ultrasound demonstrated normal thyroid gland in the midline neck. The mass was excised completely. Histopathological evaluation demonstrated mucin and fat collections with surrounding fibrovascular connective tissue (Fig. 2). Diagnosis was consistent with fibrolipoma. The patient had no periop-

erative problems and is without evidence of recurrence at a 12-month follow-up period.

3. Case 2

An eight-month-old female infant presented with a 6-month history of a right lateral tongue mass. She had no airway symptomatology and no feeding difficulties. Examination revealed a 2.0-cm firm, non-compressible bluish mass, incorporated in the intrinsic tongue musculature. There were no overlying mucosal changes. Magnetic resonance imaging of the head and neck demonstrated a well-circumscribed 1.0×2.0 cm lingual mass, which enhanced with contrast. It was hypointense on T₁ and had non-homogeneous intensity on T₂ weighted images (Fig. 3). The mass was excised transorally. Histopathological evaluation revealed atrophic muscle fibers interspersed with spindle cells and fibrous tissue (Fig. 4). Diagnosis was consistent with fibromatosis. The patient had no perioperative problems and is without evidence of recurrence at a 16-month follow-up period.



Fig. 1. Mucosalized pedunculated mass.

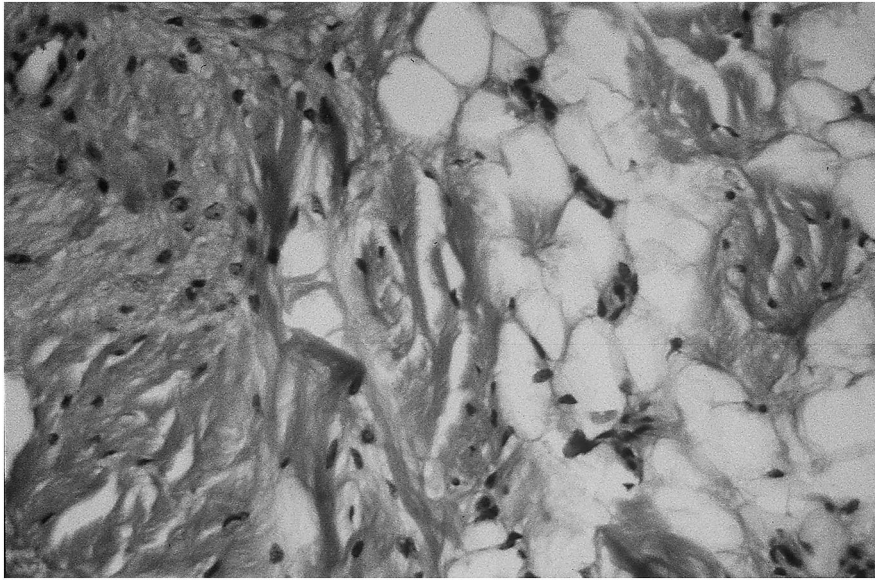


Fig. 2. Mucin and fat collections (fibrolipoma) (H&E \times 470).

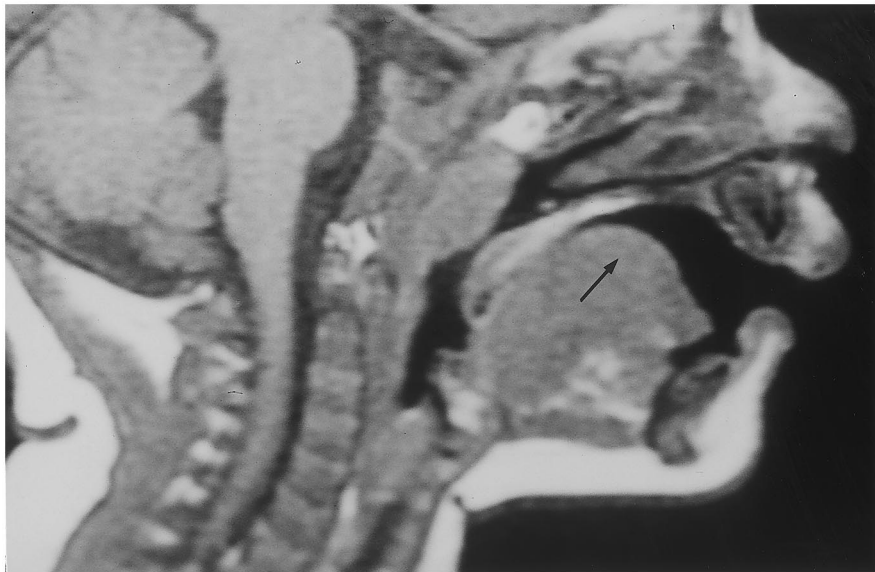


Fig. 3. Sagittal view MRI (T_2) with lateral tongue mass (arrow).

4. Case 3

A two-year-old female presented with a tongue base mass of unknown duration. She had no airway symptomatology and no interference with oral intake. Examination revealed a 1.0-cm well-

mucosalized midline tongue mass at the level of the foramen cecum. Thyroid scan demonstrated normal technetium 99m pertechnetate uptake in the region of the midline cervical thyroid gland, with no evidence of uptake in the region of the tongue base. Magnetic resonance imaging re-

vealed a cystic tongue base mass, which was of low intensity on T_1 and high intensity on T_2 . It did not enhance with contrast (Fig. 5). The mass was excised transorally. Histopathological evaluation demonstrated a cystic structure lined with nonker-

atinized squamous epithelium surrounded by skeletal muscle (Fig. 6). Diagnosis was consistent with a benign squamous epithelial cyst. She had no perioperative problems and is without evidence of recurrence at an 18-month follow-up period.

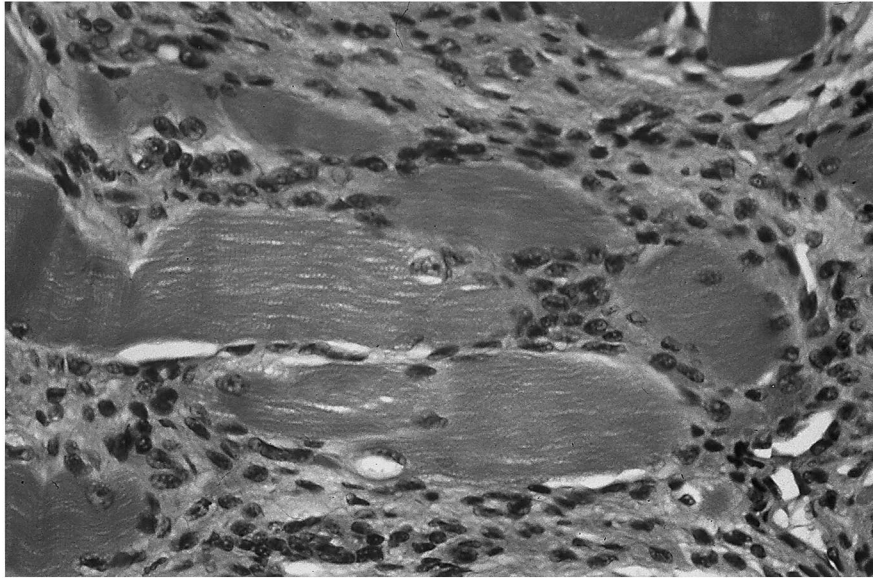


Fig. 4. Atrophic striated muscle fibers with surrounding spindle cells and fibrous tissue (fibromatosis) (H&E $\times 470$).

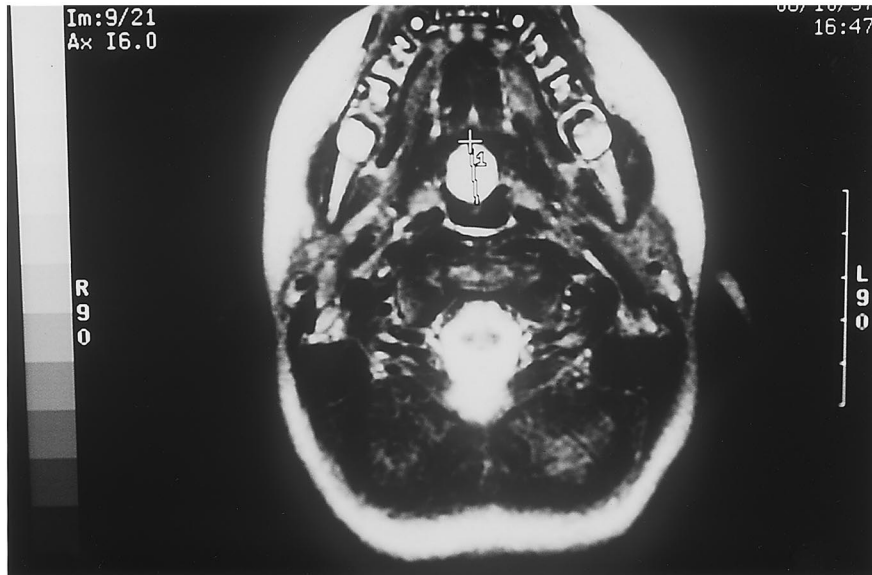


Fig. 5. Axial view MRI (T_2) with midline tongue base mass.

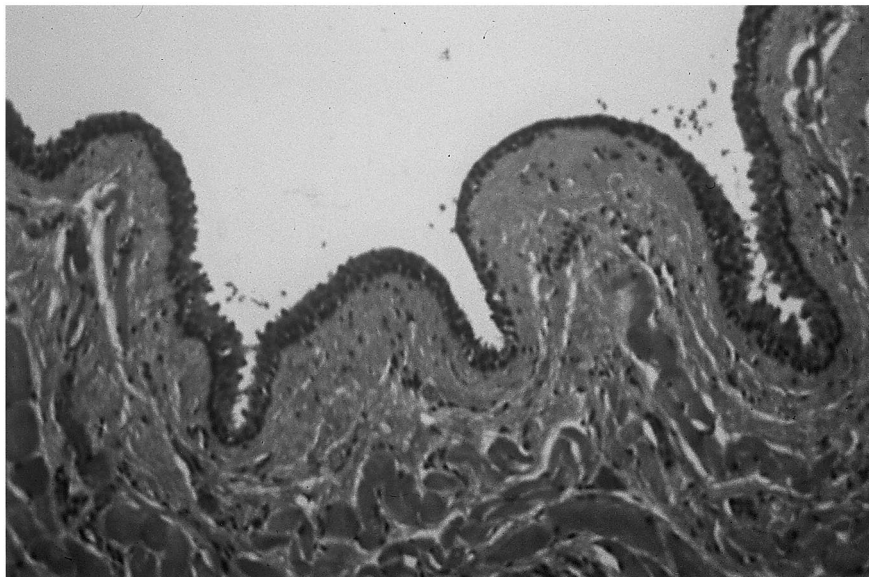


Fig. 6. Cyst wall with non-keratinized stratified squamous epithelium (squamous epithelial cyst) (H&E \times 100).

5. Discussion

Lingual tumors in the pediatric population are unusual and are predominantly benign. While they are rare in and of themselves, the tongue is the most common site of oral and maxillofacial soft tissue tumors in children. Hemangiomas are the most common pediatric oral lesion and these are most often seen on the tongue and lips [2]. Other common lingual neoplasms include lymphatic malformations, papillomas, and fibromas [2]. Malignancies of the tongue are extremely rare in children. Children who are treated with prolonged immunosuppression, or have had radiation or carcinogen exposure, are at higher risk for lingual malignancy. There may also be an association with chromosomal aberrancies associated with lingual squamous cell carcinoma [3].

The patient in Case 1 had a fibrolipoma. Lipomas and fibrolipomas are common lesions in the head and neck, but most often present in the neck as subcutaneous fatty tumors in adults. Oral lipomas are rarely seen and even less so in children. Pathology demonstrates a proliferation of fibrous and lipomatous tissue. These lesions must be completely excised and monitored for recurrence. There have been documented cases of malignant

degeneration to liposarcoma several years after excision of a benign lipoma of the neck [4].

The patient in Case 2 was found to have fibromatosis. These lesions are most commonly seen in the head and neck in both children and adults. They tend to be locally invasive due to their infiltrating fibrous proliferations. Because of their infiltrative behavior, they are considered to be intermediate between benign fibrous lesions and fibrosarcomas. Because of this behavior pattern, patients with these lesions must undergo close follow-up to monitor for recurrence [5].

The patient in Case 3 was noted to have a benign squamous epithelial cyst. These lesions develop secondary to mucus extravasation from a duct of a seromucinous gland. Given the location of this lesion at the level of the foramen cecum, the primary diagnosis from which to differentiate is a thyroglossal duct cyst. There has been much dispute regarding the appropriate radiologic work-up to evaluate for a thyroglossal duct cyst in a patient with a midline neck or tongue mass. Controversy concerning thyroid scanning, thyroid ultrasound and magnetic resonance imaging to identify presence of cervical thyroid continues to be a subject of much debate [6].

Preoperative imaging by CT or MRI scanning should be performed in several situations. If there is a question that the mass extends into the deep tongue musculature, preoperative imaging can define the depth of the mass in relation to the normal tongue tissue. If there is a question that the mass may be a hemangioma or vascular malformation, MRI imaging can elucidate these masses based on vascularity seen on MRI. All vascular lesions, except for lymphatic malformations, are isointense on T₁ weighted images and have increased signal intensity on T₂ weighted images. Lymphatic malformations may demonstrate mixed intensity on T₁ or T₂ weighted images, with absence of flow-voids [7,8]. Fast-flow lesions, such as arteriovenous malformations, will have an abundance of flow voids on MRI. Angiography would be indicated if there is consideration that the lesion is a vascular malformation. This will most clearly identify feeding vessels [9]. If the lesion is relatively superficial, does not appear to impact on lingual function or on the child's airway, and does not appear to be vascular in nature, preoperative imaging may be foregone. If there is concern that the lingual mass may be thyroid tissue, one may obtain thyroid ultrasound or thyroid scintigraphy to evaluate presence and/or function of cervical thyroid tissue [6].

Surgical excision is the mainstay of treatment for these lesions. Approach to resection is based on accessibility to the mass, size of the mass, and concern for postoperative airway obstruction secondary to lingual swelling. Most benign, non-vascular lesions of the tongue can be excised intraorally with primary closure and immediate postoperative extubation. Larger lesions may require midline mandibulotomy for exposure, with concomitant tracheotomy tube placement to bypass any potential airway obstruction. Hemangiomas may be treated by numerous methods, including observation, steroid therapy, surgical excision, laser ablation, or interferon therapy [9,10]. Lymphatic malformations may be treated in staged resections via surgical excision or laser ablation [11]. Arteriovenous malformations are treated with preoperative angiography with feeding vessel em-

bolization followed by prompt surgical resection [9].

Recurrence of benign, non-congenital tongue masses is rare if they are completely excised. However, in cases where pathology demonstrates potential for malignant degeneration (Case 1) or local invasion (Case 2), long-term follow-up is recommended.

6. Conclusion

We present three pediatric patients with benign lingual neoplasms which were not congenital in origin. Lingual tumors are unusual in children. Preoperative imaging studies aid in surgical planning and airway management. Several of the tumors have a propensity for recurrence. Close follow-up is indicated for these lesions.

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