International Journal of Pediatric Otorhinolaryngology Extra 7 (2012) 122–125

Contents lists available at SciVerse ScienceDirect

International Journal of Pediatric Otorhinolaryngology Extra

journal homepage: www.elsevier.com/locate/ijporl

Case Report

Infant tongue lesions: A case presentation and review of the literature

Jacob B. Hunter M.D.*

Montefiore Medical Center, Otorhinolaryngology, 3400 Bainbridge Avenue, Medical Arts Pavilion Third Floor, Bronx 10467, United States

ARTICLE INFO

Article history:
Received 17 February 2012
Received in revised form 12 April 2012
Accepted 14 April 2012
Available online 14 May 2012

Keywords:
Hemangiomas
Lymphangiomas
Fibromatosis
Rhabdomyosarcomas

ABSTRACT

Purpose: A case presentation of a soft tissue lesion in an 8-month-old patient's left lateral tongue and review of the literature regarding infant tongue lesions. *Methods*: The patient's medical records were reviewed with summarization of care to date that care was received at presenting institution. A systematic literature review was then conducted using PedMed. *Results*: The differential diagnosis of infant tongue lesions is large, encompassing hemangiomas, lymphangiomas, thyroglossal duct cysts, teratomas, choristomas, infantile fibromatosis and rhabdomyosarcomas, among many others. *Conclusion*: Most infant tongue lesions are benign. However, given the aggressive propensity of infantile fibromatosis and rhabdomyosarcomas, all infant tongue lesions must be characterized.

1. Introduction

The patient was born April 2010 via a normal spontaneous vaginal delivery at 40 weeks, but required subsequent hospital admission due to elevated bilirubin levels. He was then discharged home on day of life three without any sequelae. At 8 months of age the patient presented to a pediatric clinic with a cyst like lesion on the left side of his anterior tongue, noted to protrude about 1 cm, and characterized as pink and reddish in color. At that time, Mom denied any fussiness, appetite changes, fevers, recent illnesses or other lesions in the patient. He was then referred to the pediatric otolaryngology clinic.

At 11 months age the patient presented to the otolaryngology clinic, with Mom having reported that the lesion had grown in the previous 3 months, and was causing feeding issues. Mom denied any discharge from the lesion, as well as any fever, weight loss, appetite, hemoptysis or dysphagia. On clinical examination, the lesion was noted to be 1.5–2 cm in size, with no changes in color when compressed, and no discharge. The patient was then scheduled for excisional biopsy at 12 months of age.

2. Results

The specimen was noted to be a rubbery, tan and pink nodular mass, measuring 2.5 cm × 1.0 cm × 1.4 cm in its greatest dimension. On sectioning, the lesion was characterized as homogenous, tan and white in color, with a slightly fibromyxoid appearance. Pathology concluded that the mass was a pleomorphic neoplasm highly suggestive of embryonal rhabdomyosarcoma. The tumor was described as well-circumscribed, but extended into the surrounding skeletal muscle, focally to the resection margins. Mitoses were present, but not frequent, and some were atypical. There was no necrosis. Margins were involved by sarcoma, but anaplasia was not identified. Immunohistochemistry confirmed the diagnosis of embryonal rhabdomyosarcoma.

Given the diagnosis of embryonal rhabdomyosarcoma, the patient was transferred to an outside institution for chemotherapy. At the time of transfer, the patient's staging was unknown since imaging had not yet been obtained, but it was later learned with the location of metastases, that the patient was classified as Stage III.

3. Discussion

The differential diagnoses for pediatric tongue lesions are broad and greatly depend on their location in the tongue. The literature was reviewed to provide a summarization of the various pediatric oral cavity lesions, with a focus of lesions that occur either in the tongue or on the floor of the mouth in infant age children. In terms of their origin, when seen in infants, the lesions are most commonly congenital in origin [1]. It should further be noted that though these lesions may be asymptomatic, infants and children may present with symptoms of obstruction, hemorrhage secondary to trauma, and or parents concerned about cosmesis.

The tongue develops from a mesenchymal swelling in the floor of the primitive pharynx, known as the tuberculum impar. Two lateral swellings on both sides of the tuberculum impar, which grow by proliferation of the first pharyngeal arch mesenchyme, form the anterior two-thirds of the tongue. The posterior one-third of the tongue arises from the copula, which is formed by the

* Tel.: +1 3108694043.
E-mail address: jacob.b.hunter@gmail.com.

1871-4048/$ – see front matter © 2012 Elsevier Ireland Ltd. All rights reserved.
http://dx.doi.org/10.1016/j.pedex.2012.04.001

mesoderm of the second and cranial portion of the third pharyngeal arch, which fuses with the lateral lingual swellings. Thus, many lesions are found near the junction and midline of the anterior two-thirds and posterior one-third of the tongue given the tongue's embryologic origins. By separating the tongue into anterior and posterior portions, Shapiro et al. note that thyroglossal duct cysts, lingual thyroid glands, dermoids, choristomas, and heterotopic gastric cysts tend to present posteriorly and midline [1]. Horn et al. described a case series of 17 children between the ages of 1 and 132 months with tongue lesions over a 10-year period [2]. Posterior lesions included a teratoma, glial choristoma, osseous choristoma and a benign epithelial cyst [2].

In separating the tongue geographically, we first discuss the lesions that are more commonly found posteriorly. Choristomas, which are masses of histologically normal tissue in abnormal locations, can contain heterotopic islands of gastrointestinal or respiratory mucosa, have been found in the infant tongue [3]. They are also known as foregut, or enteric, duplication cysts, cystic choristomas, heterotopic gastrointestinal cysts, enterocystomas, and duplication cysts [3,4]. Capable of occurring anywhere from the oral cavity to the anus, only 0.3% have been reported in the tongue [3]. There is a male preponderance, and though some studies demonstrate a predilection for an anterior location, other studies describe a posterior predilection, there is general consensus that they are most frequently midline [4]. In terms of histology, smooth muscle is usually identified surrounding the cyst, while the cystic wall is typically composed of a mixture of columnar epithelium, which is found throughout the gastrointestinal tract, and stratified squamous epithelium [3]. The diagnosis can only be made with excisional biopsy, with three diagnostic criteria: presence of a smooth muscle coat, attachment of the cyst to a part of the alimentary tract, and the presence of mucosal lining from the alimentary tract [5]. They are thought to arise from endodermal cells, which are derived from either the stomodeum or the stomach, and are located in the neck close to the forming tongue that then become trapped during the fusion of the lateral lingual swellings over the tuberculum impar [4]. Other theories include disturbed recanalization with abnormal foregut rests forming cysts and supernumerary lung buds that may lead to cystic duplications during embryogenesis [6]. Preoperative imaging is recommended, though foregut duplication cysts are often hard to differentiate from dermoids due to the presence of proteinaceous fluid [3]. With contrast, they appear as cystic lesions that do not enhance, and appear hyper-intense on T2-weighted MRI images [3]. Surgical excision is the treatment of choice with no reported recurrences to date [3]. There have been two reported cases of malignant transformation in long-standing foregut duplication cysts of the head and neck, one case of metaplasia, and another one of adenocarcinoma [6].

Glial choristomas are lesions of normal neuroglial tissue located outside the cranium, with many theories regarding their etiology. The theories include neural tissue remnants from occipital somites that differentiated into tongue muscles, neural tissue displaced during embryogenesis, pleuripotent cell remnants in the tongue, and an extracranial variant of encephalocele [2]. As for osseous choristomas, they are lesions of normal bone tissue within soft tissue of the skin or mucosa thought to result from ossified remnants of branchial arches, which are normally located posteriorly when present in the tongue [2]. There is an increased frequency for the lesions to present during the third and fourth decades of life, with a slight female predilection [2]. For both glial choristomas and osseous choristomas, surgical excision is the treatment of choice, with no reported recurrences or malignant transformations [2].

Teratomas are the most common extragonadal germ cell tumors of childhood, involving the head and neck in 1 of every 40,000 births [7]. They contain ectodermal, endodermal and mesodermal tissue elements, with neuroepithelial tissue being the most common, and have been known to metastasize to the cervical nodes, lungs, and liver [7]. Lingual teratomas mostly occur at the foramen cecum, and are thought to arise due to the differentiation of multipotential cells sequestered during closure of the anterior neuropore or entrapment of embryonic epithelial cells along lines of first and second branchial arch closure during tongue embryogenesis [2]. Given their ability to metastasize, surgical excision is recommended.

Epithelial cysts are formed by the invagination and cystic expansion of the epidermis, which are filled with keratin and lipid-containing debris derived from sebaceous secretions [8]. They are usually dermal or subcutaneous, well-circumscribed, firm, and often moveable nodules [8].

Other lesions usually found midline and posterior include thyroglossal duct cysts and lingual thyroids, which form when ectopic thyroid tissue forms a mass in the midline at the base of tongue. The lesions have pseudostratified ciliated or squamous epithelium, with mucous glands and thyroid follicles in the adjacent stroma [7]. Occasionally, if thyroid follicles are not seen, they are termed vallecular cysts by pathologists [7]. Infants typically develop signs and symptoms within the first few weeks of life, which include stridor, feeding difficulties, failure to thrive, and breath-holding spells [7]. Lingual thyroids are noted to arise anywhere between the epiglottis and foramen cecum, while both lesions are believed to result from failed descent of the thyroid gland during embryologic development [9]. In regards to lingual thyroids, there is a four to eight times female preponderance [9]. Diagnosis is usually made during flexible or direct laryngoscopy. If a thyroglossal duct cyst or lingual thyroid is identified, then a thyroid scan or ultrasound should be performed prior to any excision to identify other functioning thyroid tissue.

In terms of anterior lesions, Shapiro provided a review of benign, non-congenital tongue lesions in children and noted that congenital lesions in the anterior portion of the tongue may be lymphatic malformations or hemangiomas [1]. In Horn et al. review, anterior lesions included a squamous papilloma, mucous cyst, fibroepithelial polyp, hamartoma, vascular malformation and a cavernous hemangioma [2].

Many studies demonstrate that hemangiomas are the most common pediatric oral lesion, routinely found on the tongue and lips [1,10]. There are several types of hemangiomas, including congenital and lobular hemangiomas, with hemangiomas of infancy (HOIs) being the most common. They are a benign proliferation of endothelial cells that are thought to be related to neural crest cells in embryologic prominences that are absent at birth, but appear and grow rapidly during the first year of life, and then undergo a period of involution [7,11]. Studies demonstrate a female preponderance, with the lip as the most frequent location for oral hemangiomas [11]. The diagnosis of HOI is usually made clinically, however further confirmation can occur via molecular staining and imaging [7]. Once identified, surgical excision is one option, but corticosteroid injection, embolization, and intralesional injections of sclerosing agents have been used to reduce the size of the lesions pre-operatively and or result in their involution. Furthermore, pulsed dye lasers have been used to reduce and remove skin redness [7].

Sako et al. discuss a case series of 250 oral and maxillofacial tumors in children under 15 years of age, reviewing their incidence and diagnosis. In children less than 1 year of age, accounting for 24 patients, the second most common oral lesions were lymphangiomas, accounting for seven of the 24 patients [10]. Studies demonstrate that 50–75% of lymphangiomas are located within the head and neck, which are congenital vascular malformations of the lymphatic system that histologically consist of abnormally