

Intralingual Dermoid Cyst in an Infant Presenting Swallowing and Sleeping Difficulties

Stefanos Dimtsas * / Nadia Theologie-Lygidakis ** / Ioannis Iatrou ***

Dermoid cysts of the oral cavity are extremely rare. The most common site is the floor of the mouth whereas intralingual location is the most unusual. They may be congenital or acquired and according to their histological appearance they are distinguished in "true" dermoid, epidermoid or teratoid cysts. We present the clinical and radiographic findings of a large congenital intralingual "true" dermoid cyst in a 10-month-old boy. The large size of the lesion and the subsequent enlargement of the tongue caused difficulties in swallowing and sleeping, symptoms which subsided after the surgical treatment. The uncommon location, the large size and the very young age of the patient were the noteworthy parameters.

Keywords: Dermoid cyst, intralingual cyst, cysts of the tongue, children.

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INTRODUCTION

Dermoid cysts are rare benign developmental lesions most often appearing in young adults and rarely in infants.^{1,2,3} They may be found, in descending order, in the gonads, the superior mediastinum and the head and neck area (7% of all dermoid cysts).

Dermoid cysts may be a) congenital, deriving mainly from ectoderm remnants entrapped during the midline fusion of the first and second branchial arches and accounts for 15% of reported cases or b) acquired, deriving from iatrogenic or traumatic implantation of epithelial cells.^{4,5,6,7} Three histologic types of the dermoid cyst are described; the Epidermoid, a keratin filled cyst lined by stratified squamous epithelium with a fibrous wall but lacking skin deriving structures, the 'true' dermoid cyst, an epithelium-lined cyst with structures such as sebaceous elements, sweat glands and hair follicles within the cyst lining and the Teratoid cyst, a cystic form that contains derivatives of endoderm, mesoderm and ectoderm within the cyst lining.^{2,3,5,7,8}

In the oral cavity, where they account for nearly 2%, der-

moid cysts are usually located in the floor of the mouth in close relationship with mylohyoid muscle^{4,5,6} and rarely in the tongue. When found in the tongue, they present as a midline swelling, which, depending on the size, may cause difficulties in swallowing, speech and sleep.^{1,2,3} A malignant transformation of oral dermoid cysts, up to 5% has been reported.^{7,9}

A case of a large «true» dermoid cyst of the tongue in an infant is presented.

Case report

A ten month old baby, presenting with a painless swelling in the tongue, was referred to the department of Oral and Maxillofacial Surgery. According to the parents, the swelling that had been small at birth, gradually increased in size causing swallowing and sleeping problems.

On clinical examination, under the normal mucosa, a round, elastic tumorous-like lesion of more than 2cm in diameter could be palpated within the mass of the anterior



Figure 1: Clinical appearance of the cyst

* Stefanos Dimtsas. Associate Specialist, OMFS, University Department of Oral and Maxillofacial Surgery at Children's Hospital "P. & A. Kyriakou," Athens, Greece.

** Nadia Theologie-Lygidakis. Lecturer, OMFS, University Department of Oral and Maxillofacial Surgery at Children's Hospital "P. & A. Kyriakou," Dental School, University of Athens, Greece.

*** Ioannis Iatrou. Associate Professor, OMFS, University Department of Oral and Maxillofacial Surgery at Children's Hospital "P. & A. Kyriakou," Dental School, University of Athens, Greece.

Send all correspondence to: N. Theologie-Lygidakis, 2 Papadiamantopoulou str., Athens 115 28, Greece

Tel: 0030 6932 308830

Fax: 0030 210 7215384

Email: lygidakis-nadia@ath.forthnet.gr

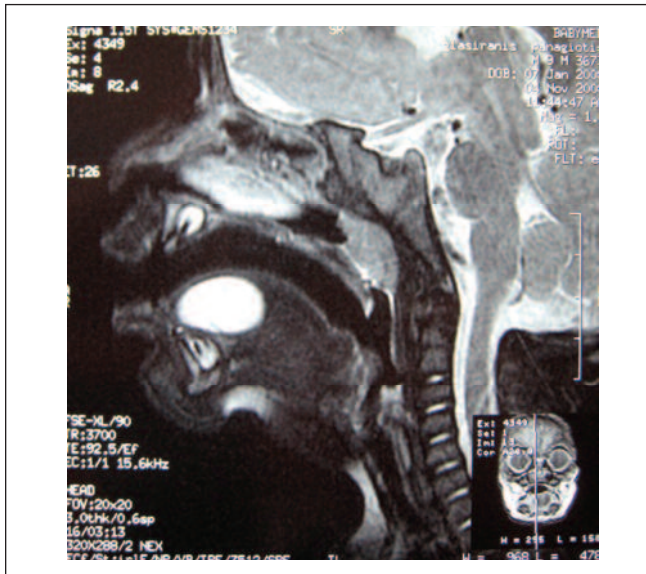


Figure 2a. MRI: a well circumscribed intralingual cystic lesion: sagittal imaging,

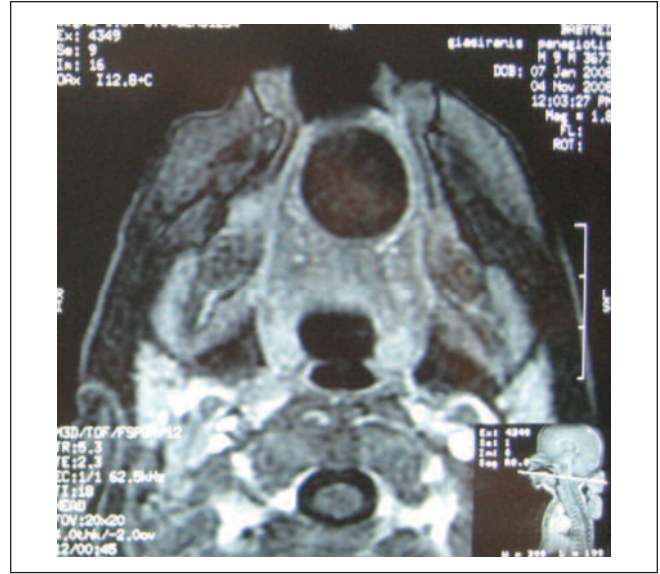


Figure 2b. MRI: axial imaging

part of the tongue (Figure 1). In MRI (magnetic resonance imaging) a well circumscribed unilocular fluid-filled cystic lesion could be observed (Figure 2 a,b).

Under nasotracheal intubation, an atraumatic incision at the ventral surface was made. The cyst was exposed, separated from the intrinsic muscles and removed in one piece (Figure 3 a,b,c). Bleeding was minimal and the wound was sutured in layers. The cyst measured 2.2x2.0x1.7cm (Figure 4). Post-operative healing was uneventful and the infant was discharged at the third day. When tongue's swelling subsided, sleeping and swallowing difficulties disappeared. According to the clinic protocols, the infant is being followed-up regularly.

Macroscopically the cyst lumen was found to contain turbid fluid with some floating whitish fragments. On histological examination, the cyst wall was found to be thin and membranous, lined by keratinized stratified squamous

epithelium and contained skin appendages, in particular sebaceous glands. Findings were compatible with “true” dermoid cyst (Figure 5).

DISCUSSION

In the 2000-2008 period, 48 patients, from the ages of 5 months to 14 years old, with oral soft tissue cystic lesions, were treated at the same department; 4 dermoid cysts, (8.3%), were excised and only one of them was intralingual dermoid cyst (2%). This percentage is in agreement with the literature.^{3,8}

Since both clinical and radiographic appearance of the case presented, were not pathognomonic, initial differential diagnosis included cystic lesions such as mucocele, ranula, thyroglossal duct cyst, lymphoepithelial cyst, lymphangioma, hemangioma and even abscess.^{1,2,4,10} The cystic nature of the lesion was verified histologically and its removal in

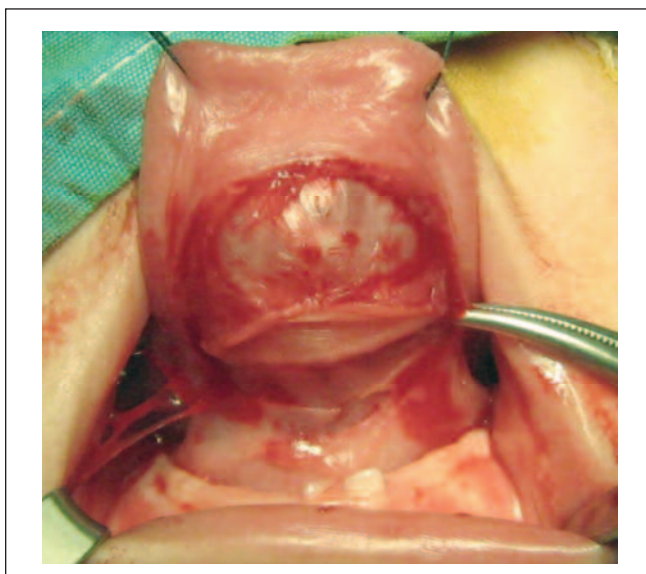


Figure 3a. Cyst exposure

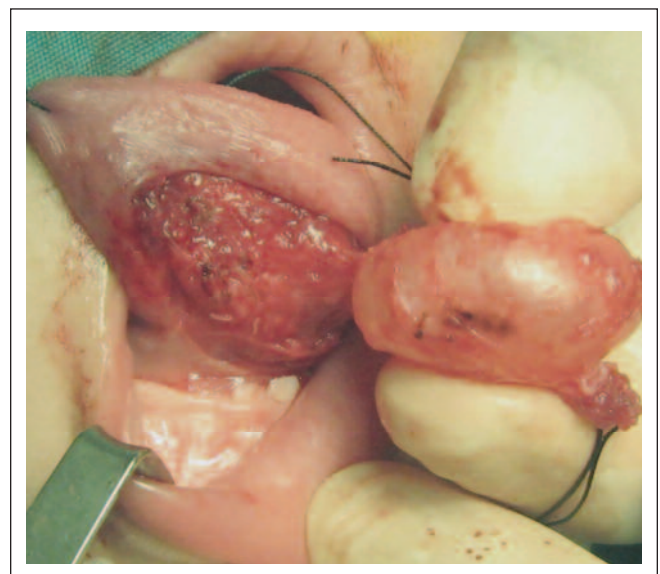


Figure 3b. Complete cyst removal

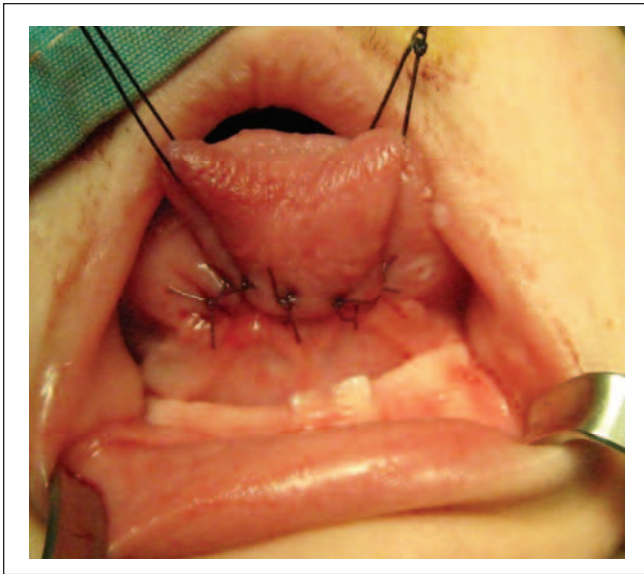


Figure 3c. Suturing

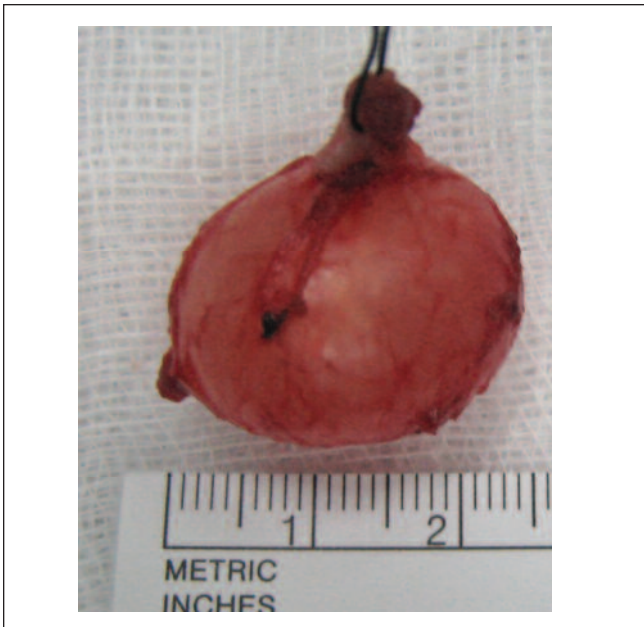


Figure 4. The specimen

one piece without any spillage was essential.

Because recurrence is possible, attributed to the fibrous capsule surrounding the cyst¹⁰ or even a malignant transformation of oral dermoid cysts up to 5%,⁹ follow-up of these cases is essential.

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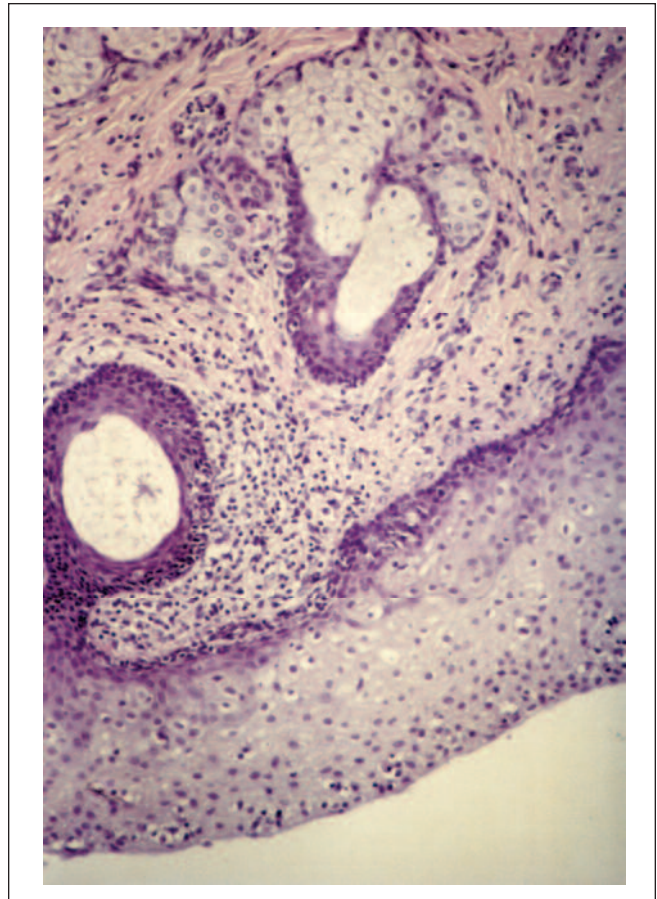


Figure 5: The cystic wall is lined by stratified squamous epithelium; presence of sebaceous glands

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