Intralingual Cystic Mass in an Adolescent with History of Hydatidosis: Case Report

Hidatidoz Hikâyesi Olan Adolesanda İntralingual Kistik Kitle

Yasin SARIKAYA,[a]
Mehmet KARATAŞ,[a]
Sedat DOĞAN,[a]
Emin KASKALAN[b]

[a]Department of Otorhinolaryngology, Adıyaman University Faculty of Medicine, [b]Clinic of Otorhinolaryngology, Adıyaman Training and Research Hospital, Adıyaman

Geliş Tarihi/*Received:* 19.12.2014
Kabul Tarihi/*Accepted:* 26.03.2015

This case report was presented as poster at 35. Turkish National Congress of ENT and Head-Neck Surgery, 2-6 November 2013, Antalya/Turkey.

Yazışma Adresi/*Correspondence:*
Mehmet KARATAŞ
Adıyaman University Faculty of Medicine, Department of Otorhinolaryngology, Adıyaman,
TÜRKİYE/TURKEY
mehmetkaratas78@gmail.com.tr

doi: 10.5336/caserep.2014-43026

ABSTRACT Lingual duplication cysts are rare in childhood and extremely rare in adulthood. The differential diagnosis of patients with complaint of mass in anterior tongue includes venolymphatic malformations, hamartoma, neurofibroma, teratoma, haemangioma, squamous cell carcinoma, glandular neoplasms, thyroglossal cyst, ranula, lingual thyroid, cyst hydatic, dermoid cyst and duplication cyst. A 16 years-old female was referred to our clinic for a mass in anterior 1/3 of tongue. She had undergone an operation for pulmonary cyst hydatic six months ago. Due to the history of pulmonary cyst hydatic, oral cyst hydatic was assumed to be the diagnosis at first sight. Differential diagnosis, imaging techniques, and treatment options for this mass are discussed.

Key Words: Tongue diseases; adolescent; echinococcosis

ÖZET Lingual duplikasyon kistleri çocukluk çağında seyrek olup erişkin yaş grubunda daha da nadir olarak görülmektedir. Dilin anterior kısmında kitle şikâyeti olan hastaların ayırıcı tanısında venolenfatik malformasyonlar, hamartoma, nörofibroma, teratom, hemanjiyom, skuamöz hücreli karsinom, glandüler neoplazmlar, tiroglossal kist, ranula, lingual tiroid, kist hidatik, dermoid kist ve duplikasyon kisti bulunur. On altı yaşındaki kız hasta dilin ön 1/3'ünde kitle nedeniyle polikliniğimize başvurdu. Hasta 6 ay önce pulmoner kist hidatik nedeniyle opere edilmiş. Bu nedenle dildeki kitlenin ön tanısı başta kist hidatik olarak düşünüldü. Bu olgu sunumunda bu kitlenin ayırıcı tanısı, görüntüleme yöntemleri ve tedavi yaklaşımları tartışıldı.

Anahtar Kelimeler: Dil hastalıkları; adolesan; ekinokokkoz

Turkiye Klinikleri J Case Rep 2016;24(1):34-8

Duplication cysts (DC) are relatively rare encountered gastrointestinal masses. They are also called as choristoma, heterotopic gastrointestinal cysts, or enterocytoma.[1,2] These cysts can be seen anywhere from oral cavity to anus and they arise mostly from small intestine. 0.3% of these cysts occur in the oral cavity and they arise relatively rare from 1/3 of anterior tongue.[3] Most of them are recognized just after birth without any symptom but when they arise in oral cavity they have potential to cause respiratory and feeding difficulties in newborn. In the literature it has been reported that DC has malign transformation potential in adults so that they should be excised as soon as possible.[4] DCs are lined with respiratory, gastric, squamous, or ciliated epithelium but mostly combination of these.[5] The differential diagnosis of adult patients with complaint of

mass in anterior tongue includes venolymphatic malformations, hamartoma, neurofibroma, teratoma, haemangioma, squamous cell carcinoma, glandular neoplasms, thyroglossal ductus cyst, ranula, lingual thyroid, cyst hydatic, dermoid cyst and DC. Radiological imaging has a crucial role for differential diagnosis and surgical intervention in these patients. In this case, a 16 years-old female was referred to our clinic for a mass in anterior 1/3 of tongue which was incidentally found in cranial magnetic resonance imaging (MRI). Interestingly, she had history of a pulmonary surgery for cyst hydatic disease six months ago. Herein, differential diagnosis, imaging techniques, and treatment options of this lingual mass are discussed.

CASE REPORT

A 16 years-old female patient referring to a neurologist with complaint of headache had lingual mass which was incidentally found in cranial MRI. In the past history, patient had noticed the mass ten years ago with no any disturbing symptom up to the time and she was operated for pulmonary cyst hydatic six months ago. Physical examination showed 2x2 cm, intralingual, smooth, painless and cystic mass in 1/3 of anterior tongue. Due to the history of pulmonary cyst hydatic, oral cyst hidatic was assumed to be the diagnosis at first sight. Cervical computed tomography (CT) revealed a 2.5 cm well-circumscribed hyperdense mass within the midline of anterior tongue with no contrast enhancement in postcontrast series (Figure 1). In cervical MRI, there was 2x1.7 cm in size, uniformly shaped, anterior intralingual hyperintense mass in all T1, T2 and fat supressed series and it was nonenhancing in post contrast series (Figure 2). There was no pathology in thyroid and the rest of neck. Chest X-rays were normal. Because of positive history for pulmonary hydatic cyst, serologic tests for hydatosis were done but found negative. Fine needle aspiration cytology (FNAC) showed brown fluid with no specific diagnosis. Differential diagnosis included venolymphatic malformations, hamartoma, neurofibroma, teratoma, haemangioma, squamous cell carcinoma, glandular neoplasms, thyroglossal ductus cyst, ranula, lingual thyroid, cyst hydatic, dermoid cyst and DC.

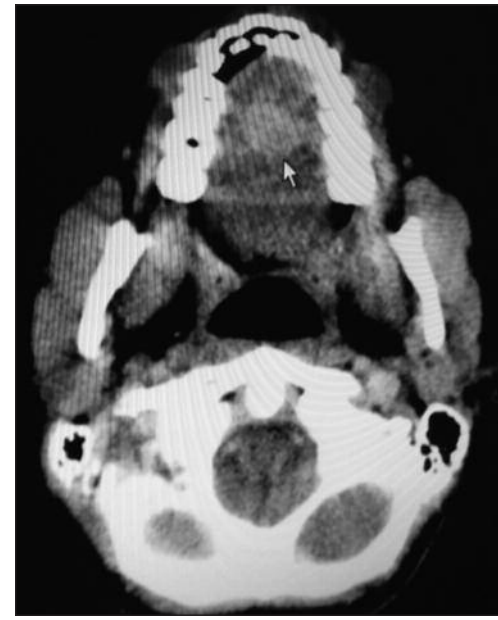

FIGURE 1: Axial CT of tongue shows well-circumscribed hyperdense mass within the midline of anterior tongue.

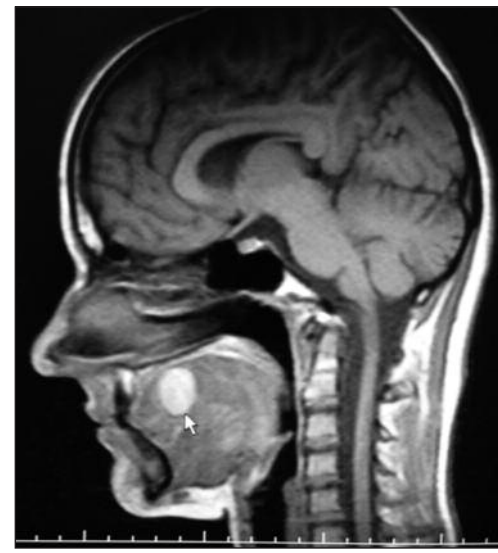

FIGURE 2: Midline sagittal T1-weighted images of tongue shows the location of the cyst within the intrinsic muscles of the tongue. The cyst is of hyperintense due to its proteinaceous content.

Under general anaesthesia with transnasal intubation a midline incision on dorsal surface of anterior tongue was done. There was a cystic mass with regular border just beneath the mucosa. It was completely excised without any spillage in the operative field (Figure 3, Figure 4). Histopathology

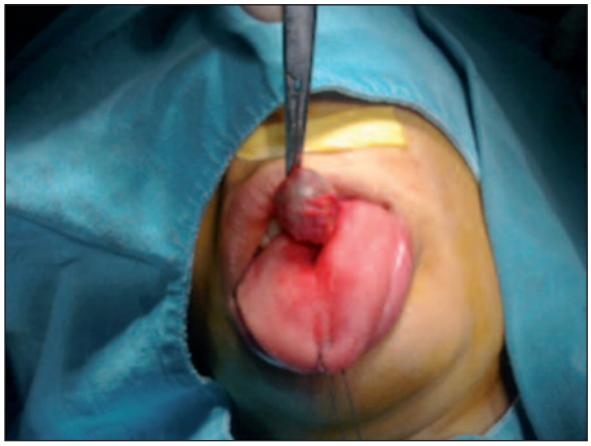

FIGURE 3: Excision of the cyst via a dorsal incision in the tongue.

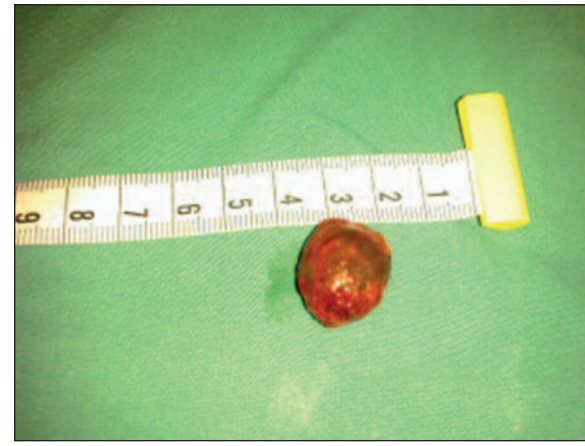

FIGURE 4: Gross appearance of the cyst.

showed duplication cyst which was lined by squamous, columnar and gastric epithelium. At postoperative day 3, patient was discharged. There was no recurrence on follow-up for 1 year (Figure 5).

DISCUSSION

Alimentary tract DCs are migrational anomalies which take place during fetal life. These cysts can be seen anywhere from oral cavity to anus and they arise mostly from small intestine. 1/3 of gastrointestinal duplication cysts occur in the foregut, and foregut duplication cysts (FDC) are classified as bronchogenic, esophageal, and neuroenteric.[5] DCs are lined with respiratory, gastric, squamous, or ciliated epithelium but mostly combination of these.[5] 0.3% of these cysts occur in the oral cavity and they arise relatively rare from 1/3 of anterior tongue.[3,6] Lingual FDCs (LFDC) do not fall into the above classification. Four main theories have tried to explain pathogenesis of LFDCs.[7] But none of them can explain incorporation of the heterotopic enteric tissue into the tongue. It was postulated that since foregut and pharyngeal arches are closely apposed LFDCs may arise from abnormal cellular migration.[8]

The clinical presentation of DCs is a reflection of its location, its mass effect, and complications of the ectopic mucosal lining.[6] Most of DCs are recognized just after birth without any symptom but when they arise in oral cavity there is a potential

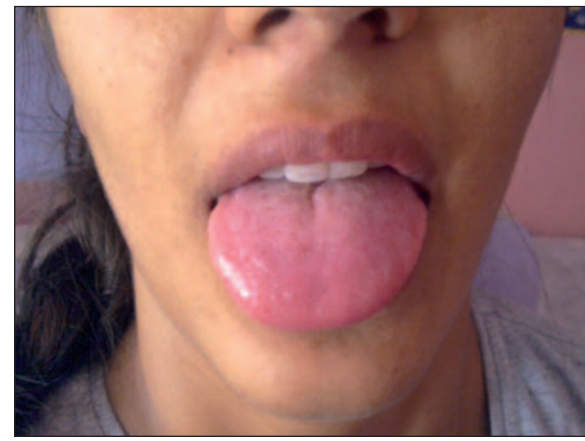

FIGURE 5: Postoperative 12th month with no any recurrence or sequela.

for respiratory and/or feeding problems in the infant. While mass effect is a common presentation in younger patients, because of DCs' malign transformation potential in adults, they should be excised as soon as possible.[4]

The differential diagnosis of a mass in the anterior tongue includes venolymphatic malformations, hamartoma, neurofibroma, teratoma, haemangioma, squamous cell carcinoma, glandular neoplasms, thyroglossal ductus cyst, ranula, lingual thyroid, cyst hydatic, dermoid cyst, and DC.[5,9-11] In this case since patient had a positive history of cyst hydatic operation six months ago, at first we presumed the mass as lingual cyst hydatic (LCH). Hydatidosis commonly appear as cystic lesions and these characteristically grow

slowly (1-2 cm/year).[12,13] MRI and CT are major tools of diagnostic imaging. Since MRI provides excellent contrast resolution of soft tissues with multiple pulse sequences, it is the technique of choice for oral cavity lesions. In the absence of dental amalgam enhanced CT can also be useful. FNAC is preferable with minimal complications.[14] Serologic tests are contentious. In our case, cervical CT revealed a 2.5 cm well-circumscribed hyperdense mass within the midline of anterior tongue with no contrast enhancement in postcontrast series. Similarly LCH usually reveals as hyperdense area on CT imaging. Cervical MRI showed 2x1.7 cm in size, uniformly shaped, anterior intralingual hyperintense mass in all T1, T2 and fat supressed series and it was nonenhancing in post contrast series. Since LCH presents as hypointense in T1, hyperintense in T2 and some enhancement in postcontrast series, MRI findings of the mass were incompatible with cyst hydatic. FNAC showed no specific diagnosis. However, unlike clear appearance in hydatosis, there was brownish appearance of the fluid. Serologic tests for hydatosis were done but found negative. Complete surgical excision of mass was performed. Histopathological exam showed DC lined by squamous, columnar and gastric epithelium. Although, in the literature, few cases of LCH were reported, in the countries like Africa, Europe, Asia, the Middle East, Central and South America, LCH should be also suspected in the patients complaining of tongue mass with or without history of hydatic cyst disease.[10,15] Since fatal reactions, like anaphylaxis, can occur, otolaryngologist should take into consideration the possibility of LCH during lingual cyst excision. Any lingual cyst should be excised completely without any spillage.

Most of DCs are recognized just after birth with potential to cause respiratory and feeding difficulties. Initial diagnosis of them is usually done with USG during fetal life in which echogenic inner mucosal layer and hypoechoic outer muscular layer can be seen.[3] MRI, with its lack of ionizing radiation and superior soft tissue resolution, is the imaging study of choice for fetus. Therefore, surgical excision and prevention of complications can be achieved safely just after delivery. But in adolescent period LDCs, especially intralingual ones, are extremely rare. They should be kept in mind in the differential diagnosis of a tongue mass. Diagnosis is made by histopathologic exam. Treatment of choice is complete excision, as soon as possible, because of malign transformation potential. There was no any recurrence reported in the literature.

In conclusion, DCs are rare encountered lesions and may be discovered accidentally during radiographic examination, body scanning, surgery, or for other clinical reasons. Therefore, otolaryngologists should be meticulous about the patient's past history and country and location, duration, and imaging features of the lingual mass. Cyst hydatic should be kept in mind in the differential diagnosis of lingual cystic masses and the potential fatal reactions, like anaphylaxis, should be taken into consideration pre- and peroperatively.

REFERENCES

1. Rousseau T, Couvreur S, Senet-Lacombe E, Durand C, Justrabo E, Malka G, et al. Prenatal diagnosis of enteric duplication cyst of the tongue. Prenatal Diagn 2004;24(2): 98-100.
2. Karam O, Pfister RE, Extermann P, LaScala GC. Congenital lingual cysts. J Pediatr Surg 2007;42(4)E25-7.
3. Kong K, Walker P, Cassey J, O'Callaghan S. Foregut duplication cyst arising in the floor of mouth. Int J Pediatr Otorhinolaryngol 2004; 68(6):827-30.
4. Volchok J, Jaffer A, Cooper T, Al-Sabbagh A, Cavalli G. Adenocarcinoma arising in a lingual foregut duplication cyst. Arch Otolaryngol Head Neck Surg 2007;133(7):717-9.
5. Patel P, Branstetter BF 4th, Myers EN. Lingual foregut duplication in a middle-aged adult. AJNR Am J Neuroradiol 2011;32(3): E40-1.
6. Azzie G, Beasley S. Diagnosis and treatment of foregut duplications. Semin Pediatr Surg 2003;12(1):46-54.
7. Eaton D, Billings K, Timmons C, Booth T, Biavati JMJ. Congenital foregut duplication cysts of the anterior tongue. Archives of Otolaryngology Head Neck Surg 2001;127(12):1484-7.
8. Manor Y, Buchner A, Peleg M, Taicher S. Lingual cyst with respiratory epithelium: an entity of debatable histogenesis. J Oral Maxillofac Surg 1999;57(2):124-7.
9. Madan HK, Swain L, Borkar J. Anesthetic management of a neonatal lingual gastric duplication cyst: report of a rare case. J Anesth 2012;26(3):438-41.

10. Zaimi A. [Hydatid cyst of the tongue]. Tunis Med 1975;53(5):303.
11. Lalwani AK, Lalwani RB, Bartlett PC. Heterotopic gastric mucosal cyst of the tongue. Otolaryngol Head Neck Surg 1993;108(2): 204-5.
12. Gangopadhyay K, Abuzeid MO, Kfoury H. Hydatid cyst of the pterygopalatine-infratemporal fossa. J Laryngol Otol 1996;110(10):978-80.
13. Senneroglu L, Onercĩ M, Turan E, Sungur A. Infratemporal hydatid cyst--unusual location of echinococcosis. J Laryngol Otol 1994;108(7): 601-3.
14. Amice J, Sparfel A, Pétillon F, Amice V, Jézéquel J, Rivière MR. Hydatid cyst of the neck: diagnosis by fine needle aspiration. Acta Cytol 1992;(36(3):454-6.
15. Perl T, Goldberg B. Hydatid cyst in the tongue. Oral Surg Oral Med Oral Path 1972;33(4):579-81.