

Chondrolipoma of the Tongue in a Child: A Case Report

Suheyl Haytoglu^{a, c}, Nur Yucel Ekici^a, Ozgur Kulahci^b, Osman Kursat Arikan^a

Abstract

Chondrolipomas are seen very rare in the oral cavity. They are benign mesenchymal tumors which are characterized by the proliferation of mature adipocytes associated with the deposition of mature cartilaginous tissue. English literatures show that these lesions are usually small (0.5 - 2.5 cm), duration of disease before diagnosis ranges from 2 months to 2 years and the peak incidence of lipomas is during the fifth and sixth decade of life. A 16-year-old boy presented with a painless mass on the right lateral border of the tongue since birth in this case report. Intraoral clinical examination revealed a medium hard, multilobular, irregular mass, consistency measuring approximately 6 × 4 cm in its major diameter. Therefore, the present case is peculiar due to its long duration (approximately 16 years) and its size.

Keywords: Chondrolipoma; Oral cavity; Tumor; Tongue

Introduction

Lipomas are common, benign, slow-growing, soft tissue neoplasms of mature adipocytes; however, only 1-4% of them involve the oral cavity [1-3]. Most of the cases are diagnosed in patients over the age of 40 and these are more commonly seen in males than females [3-5]. Oral lipomas have been reported to occur in a variety of locations including the salivary glands, buccal mucosa, gingiva, lip, tongue and floor of the mouth. It appears as a single or lobulated long-standing painless lesion with either a sessile or a pedunculated base. Different histopathological variants have been recognized, such

as fibrolipomas, angiolipomas, myolipomas, spindle cell lipomas, chondroid lipomas, chondrolipomas and osteolipomas. Among the histopathological variants, lipomas with cartilaginous metaplasia, called chondrolipomas, are uncommon in the oral cavity [3, 6]. The appearance of chondrolipomas in a child is very unusual. Here, we describe a case of chondrolipoma of the tongue in a 16-year-old boy.

Case Report

A 16-year-old boy presented with a painless mass on the middle of the right lateral border of tongue (Fig. 1). The lesion was first noticed about 15 years earlier which was a hazelnut-sized mass by his family and had not grown remarkably so far. He noticed an increase in the size of the mass over the past 1 year.



Figure 1. Clinical image shows mass originating from right lateral border of tongue.

Manuscript accepted for publication June 30, 2015

^aDepartment of Otolaryngology, Head and Neck Surgery, Adana Numune Education and Research Hospital, Adana, Turkey

^bDepartment of Pathology, Adana Numune Education and Research Hospital, Adana, Turkey

^cCorresponding Author: Suheyl Haytoglu, Department of Otolaryngology, Head and Neck Surgery, Adana Numune Education and Research Hospital, Suleyman Demirel Blv. N0:11/15, Adana, Turkey.
Email: drsuheylhayt@hotmail.com

doi: <http://dx.doi.org/10.14740/jmc2213w>

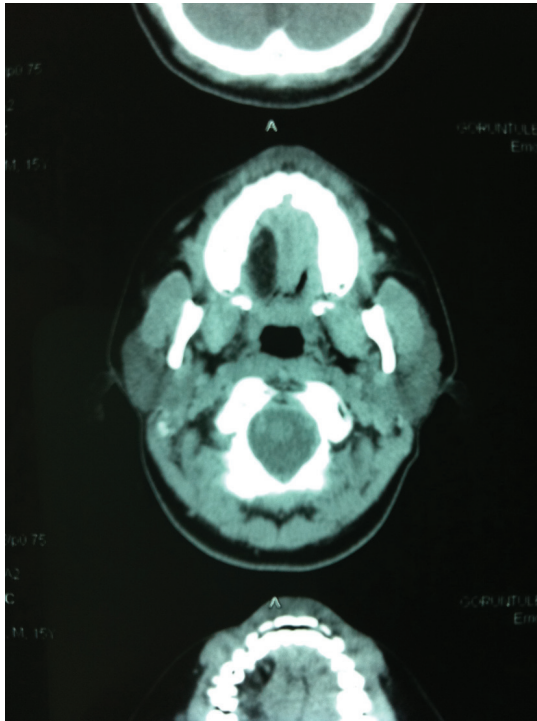


Figure 2. Radiological investigation shows a well-demarcated lesion at the lateral border of the tongue.

At the time of presentation it was causing problems with speech and eating, and was subject to repeated trauma due to accidental biting. Intraoral clinical examination revealed a medium hard, multilobular, irregular mass, consistency measuring approximately 6×4 cm in its major diameter, which was located along the midline of the right lateral border of the tongue with no evidence of submandibular and cervical lymphadenopathy. No ulceration was noted. A detailed systemic examination and laboratory investigations were within the normal limits. His family history was unremarkable. Radiological investigations showed a well-circumscribed fatty mass in the right side of the tongue (Fig. 2). Histological examination of biopsy revealed it was composed of mature adipose tissue arranged in lobules that are separated by fibrous tissue septa. Under general anesthesia, the lesion was removed totally with following dropped 1 cm strong border (Fig. 3). Microscopic examination revealed a benign neoplasm characterized by the proliferation of mature adipocytes that were separated by fibrous connective tissue and presence of a central area of mature cartilaginous tissue that exhibited lacune filled with chondrocytes (Fig. 4, 5). No evidence of malignant change was found and chondrolipoma of the tongue was diagnosed. A follow-up period of 2 years shows complete healing with no recurrence. Informed consent of the patient has been obtained.

Discussion

Lingual masses are seen very rare in children. They are predominantly benign and congenital. Available data show that



Figure 3. Clinical image shows the excised specimen.

lesions are usually small (0.5 - 2.5 cm) and duration of disease before diagnosis ranges from 2 months to 2 years. Therefore, the present case is peculiar due to its long duration (approximately 16 years) and its size (approximately 6×4 cm). The peak incidence of lipomas is during the fifth and sixth decade of life [3, 7]. Our case is a 16-year-old boy. Bures and Barnes reported that a benign mesenchymoma may represent a hamartoma if the lesion occurs in an individual younger than 25 years, associated with other congenital anomalies or hamartomatous lesions or with a phacomatosis [7]. In our case, detailed systemic examination and laboratory investigations were within the normal limits.

The pathogenesis of chondrolipomas is uncertain. Although the exact origin of the fat cells, chondroblasts remains controversial, it is generally accepted that they develop from different types of undifferentiated mesenchymal cells. The main mechanisms is differentiation of pluripotent mesenchymal cells into adipose tissue and cartilage [3, 5, 8]. Hietanen and Makinen [6] and Maes and Eulerink [9] also suggest that the tumor is essentially a lipoma or chondroma and the accompanying cartilage or fat is a form of metaplastic stromal reaction. Mesenchymal cells can be modified by local or systemic factors such as local trauma and prolonged ischemia [3, 6, 8, 10]. In our case, the lesion was first noticed about 15 years earlier which was a hazelnut-sized mass by his family and he noticed an increase in the size of the mass over the past 1 year. Nakano et al [11] suggest that the expression of transforming growth factor-beta (TGF- β), latent TGF- β binding protein-1 (LTBP-1) and bone morphogenetic protein (BMP) might be important in the pathogenesis of chondrolipoma.

Lipomas are occasionally altered by metaplastic elements

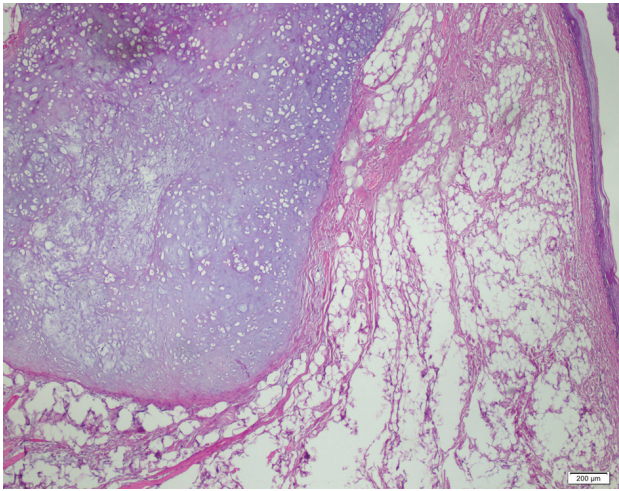


Figure 4. Histopathological image shows island of mature cartilage, surrounded by mature fat cells (H&E stain, × 40).

that form an intrinsic part of the tumor, the commonest variant being fibrolipoma [6, 10]. There is not any cartilage tissue in tongue normally. It is possible that the present case started out as either lipoma or chondroma. Exceptionally in lipomas of large size and long duration, cartilaginous metaplasia may occur. Lingual chondrolipoma or lipochondroma was first described in 1989 in a 47-year-old male who presented with a firm mass less than 1 cm in diameter at the lateral border of tongue [9].

Chondrolipomas are characterized by the proliferation of mature adipocytes associated with the deposition of mature cartilaginous tissue on histopathological examination [3, 6]. Despite the similar nomenclature, chondrolipomas are different from chondroid lipomas. Chondroid lipomas are characterized by the proliferation of mature adipocytes arranged in the extracellular matrix with chondroid, myxoid and hyalinized areas [3, 5]. In contrast to chondroid lipomas, chondrolipomas do not contain lipoblast-like cells and are easily identified as benign tumors [3, 10]. In addition, chondroid lipomas can be differentiated from myxoid liposarcomas and extraskeletal myxoid chondrosarcomas by the lack of nuclear atypia, mitotic activity, characteristic vascular pattern, and the presence of both chondroid and fat cells [3, 12]. The treatment of these tumors consists of surgical excision, with no cases of recurrence having been reported. Chondrolipomas are well-demarcated tumors and can therefore be easily removed. Chondrolipoma shows a clear separation between the cartilaginous component and the fatty component, which makes the appearance of this lesion more benign.

Other differential diagnosis of chondrolipomas includes osteochondrogenous choristomas and malign tumors. Osteochondrogenous choristomas differ from chondrolipomas by the formation of abundant bone or cartilage, despite poor fat tissue, upon microscopic examination. Chondroid lipomas can be differentiated from chondrolipomas by the lack of lipoblast-like features. Malignant tumors can be distinguished from chondrolipomas because they usually present cytologic features of malignancy such as mitotic activity and nuclear atypia [3, 8, 13].

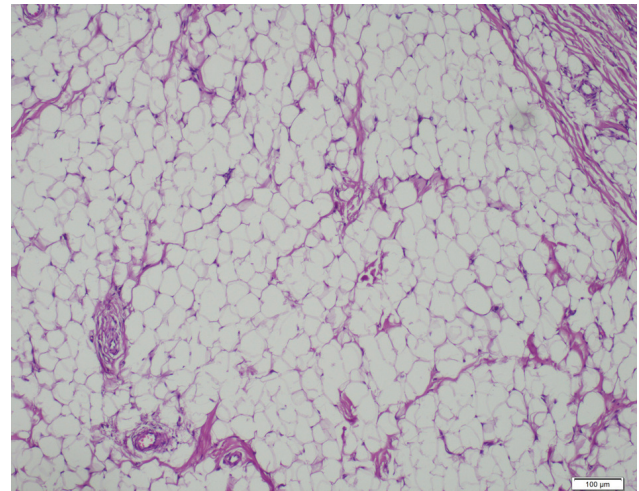


Figure 5. Well-demarcated proliferation of mature fat cells arranged in lobules separated by connective fibrous tissue septa.

The treatment of choice for these tumors consists of surgical excision. They are well-demarcated tumors and can therefore be easily removed. After complete surgical excision no recurrence was described [1, 4, 7, 14].

In conclusion, chondrolipoma was rarely seen in tongue especially in childhood. The pathogenesis is inconclusive. Although it is a benign tumor, a complete surgical excision must be done because of its localization and potential complications.

Conflicts of Interest

There are not any potential conflicts of interest.

References

1. Manor E, Sion-Vardy N, Joshua BZ, Bodner L. Oral lipoma: analysis of 58 new cases and review of the literature. *Ann Diagn Pathol.* 2011;15(4):257-261.
2. Fregnani ER, Pires FR, Falzoni R, Lopes MA, Vargas PA. Lipomas of the oral cavity: clinical findings, histological classification and proliferative activity of 46 cases. *Int J Oral Maxillofac Surg.* 2003;32(1):49-53.
3. Nonaka CF, Miguel MC, de Souza LB, Pinto LP. Chondrolipoma of the tongue: a case report. *J Oral Sci.* 2009;51(2):313-316.
4. Tasic D, Pavlovic M, Stankovic D, Dimov I, Stanojevic G, Dimov D. Ossifying chondrolipoma of the tongue. *Vojnosanit Pregl.* 2012;69(11):1009-1012.
5. Furlong MA, Fanburg-Smith JC, Childers EL. Lipoma of the oral and maxillofacial region: Site and subclassification of 125 cases. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod.* 2004;98(4):441-450.
6. Hietanen J, Makinen J. Chondrolipoma of the tongue. A case report. *Int J Oral Maxillofac Surg.* 1997;26(2):127-128.
7. Goel G, Khadilkar UN, Kumar S. Chondrolipoma of

- tongue. *Kathmandu Univ Med J (KUMJ)*. 2008;6(24):505-507.
8. Fujimura N, Enomoto S. Lipoma of the tongue with cartilaginous change: a case report and review of the literature. *J Oral Maxillofac Surg*. 1992;50(9):1015-1017.
 9. Maes A, Eulderink F. Chondrolipoma of the tongue. *Histopathology*. 1989;14(6):660-662.
 10. Weiss S, Goldblum JR, editors. Benign lipomatous tumors. In: *Enzinger and Weiss's soft tissue tumors*, 4th ed. Mosby, St Louis, 2001. pp. 571-639.
 11. Nakano M, Arai E, Nakajima Y, Nakamura H, Miyazono K, Hirose T. Immunohistochemical study of chondrolipoma: possible importance of transforming growth factor (TGF)-betas, latent TGF-beta binding protein-1 (LTBP-1), and bone morphogenetic protein (BMP) for chondrogenesis in lipoma. *J Dermatol*. 2003;30(3):189-195.
 12. Darling MR, Daley TD. Intraoral chondroid lipoma: a case report and immunohistochemical investigation. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod*. 2005;99(3):331-333.
 13. Berg T, Gorsky M. Chondrolipoma of the tongue: a case report of a rare diagnosis. *Int J Dermatol*. 2010;49(4):441-442.
 14. Batchvarova Z, Kadlub N, Coulomb-L'Hermine A, Picard A, Galliani E. Giant chondrolipoma of the tongue in a 14-year-old child with mandibular hypertrophy. Isolated lesion or regional overgrowth syndrome. *Int J Oral Maxillofac Surg*. 2012;41(2):261-264.