Int. Adv. Otol. 2010; 6:(1) 120-123

CASE REPORT

Osteolipoma of the External Ear Canal

Abdullah Durmaz, Bulent Kurt, Hakan Birkent, Yusuf Hidir, Mustafa Gerek

Department of Otorhinolaryngology Head and Neck Surgery, Gulhane Military Medical Academy, Etlik 06018, Ankara-Turkey.

A case of osteolipoma in the external ear canal was reported. A twenty-one-year-old male patient was referred for a mass in the left external ear canal causing mild fullness and hearing loss. Radiological evaluation was doubtful for osteolipoma. The well-demarcated, firm mass was excised via postauricular approach, and histopathologic diagnosis of osteolipoma was made. To our best knowledge, this is the first report of osteolipoma of the external ear canal in the available literature. This case indicates that osteolipoma should be kept in mind in the differential diagnosis of external ear canal mass as an extremely rare benign lesion. The case report and a review of the literature concerning osteolipoma of the external ear canal and the head and neck are presented.

Submitted : 27 May 2009 *Revised : 20 November 2009* *Accepted : 13 December 2009*

Introduction

Lipomas are the most common soft tissue tumors of adulthood in any location of the body, and they are subclassified according to particular morphologic features [1]. Lipomas displaying osseous structures are referred to as osteolipomas or ossifying lipomas [2]. Osteolipomas are very rare and usually located in the intraosseous regions or adjacent to the bone tissue [3]. Most of the lipomas that contain osseous tissue are located in long bones (intraosseous) or adjacent to bones (parosteal). Parosteal osteolipomas are exceedingly rare and interestingly most of them occur in the head and neck region. Although the osteolipomas have been reported in the head and neck region, to our knowledge, the case presented herein is the first case of osteolipoma in the external ear canal.

Case Report

This is a 21-year-old male who was referred for a mass in the left external ear canal. The patient had first noted the lesion in childhood. It had grown continually and caused mild fullness and hearing loss on the left ear. He had no pain or any other signs of mass. He had no remarkable past or family history.

On physical examination, the mass was hard, well demarcated, approximately 1 cm in diameter and located in the entrance of the left external ear canal. It was bony hard and immovable on palpation, impeding to otoscopic examination for medial part of the canal and ear drum. Audiologic examination showed mild conductive hearing loss in pure tone audiometry. Tympanometry could not be performed appropriately. No other abnormality was found in the head and neck and a general medical evaluation was normal.

Computed tomography (CT) of the temporal bone revealed a well demarcated mass originating from the posterior wall of the left external ear canal entrance (Figure 1). There were soft tissue and bony density elements within the mass and clearly showed a dense osseous cover. The left middle ear, inner ear and the mastoid cells were normal. Magnetic resonance imaging (MRI) of the temporal bone demonstrated a mass which had heterogenic signal intensity on T1 and T2-weighted images in location as described on CT. The mass had suppressed signal intensity on fat-suppressed images. The cortical bone cover appeared to be a hypointense line around the mass (Figure 2). Probable radiological diagnosis indicated a benign lesion like osteochondroma.

Corresponding address:
Abdullah Durmaz, MD, Assistant Professor
Dept. of ORL & HNS, GATA Etlik 06018, Ankara Turkey
Phone: +90 (312) 304 5701; Fax: +90 (312) 304 5700; E-mail: adurmaz@gata.edu.tr

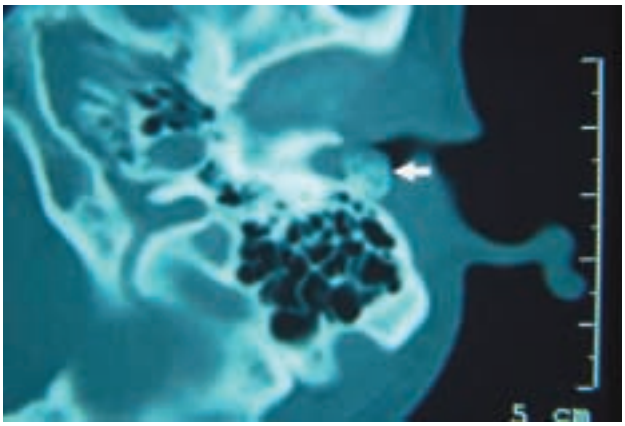

Figure 1. Axial CT image of the temporal bone. The osteolipoma (arrow) originating from the posterior wall of the left external ear canal displays soft tissue and bony density elements within and a dense osseous cover.

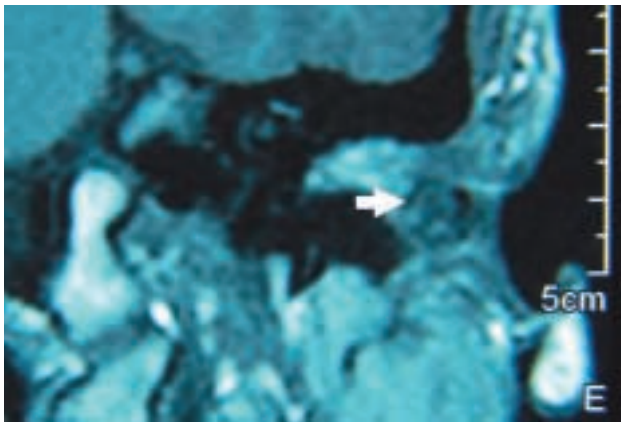

Figure 2. Coronal T2-weighted MR image of the osteolipoma. The dense osseous cortical bone cover shows a hypointense cortical line circumscribing a fatty core (arrow).

Postauricular approach was employed for mass resection under general anesthesia. The mass was feebly attached to the temporal bone with an osseous pedicle (Figure 3). It was completely excised by blunt dissection and attachment point to the cortical bone was shaved with a diamond burr. Gross examination showed a firm smooth mass measuring 0.8x1.2 cm. On microscopy, the tumor consisted of mature adipose tissue and fibrous tissue covered by a thin lamellar bone (Figure 4). Bony trabeculae showed focal osteoblastic and osteoclastic activity. Postoperative course was uneventful, and the hearing loss improved.

Discussion

Twenty cases of osteolipomas have been reported in the head and neck region. These are located in oral cavity [4-9], oropharynx [10], nasopharynx [11], sinonasal [12], frontal [13], maxillo-facial [14], temporo-parietal [15], skull base [16], parapharyngeal [17-19], retropharyngeal [20], submandibular [21], and neck [3,22]. To our knowledge, the case presented herein is the first case of osteolipoma located in the external ear canal. and it is possibly a parosteal osteolipoma because of the localization and its adhesion to surrounding bone with an osseous pedicle.

Although osteolipomas are benign slow-growing tumors and usually asymptomatic painless masses in the head and neck. As they get bigger, they can

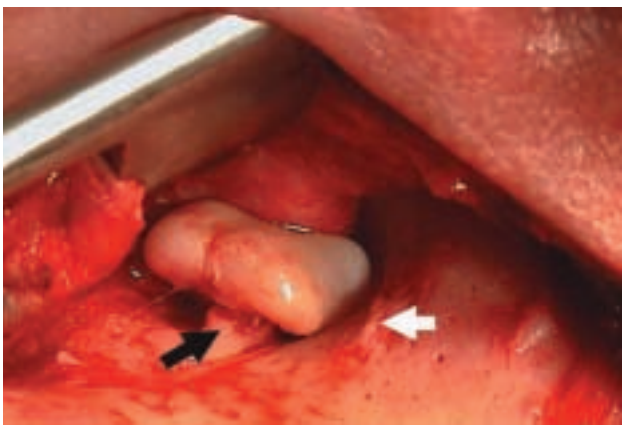

Figure 3. An intraoperative appearance of the osteolipoma. A: auricula, black arrow: osseous pedicle of mass, white arrow: Henle spine.

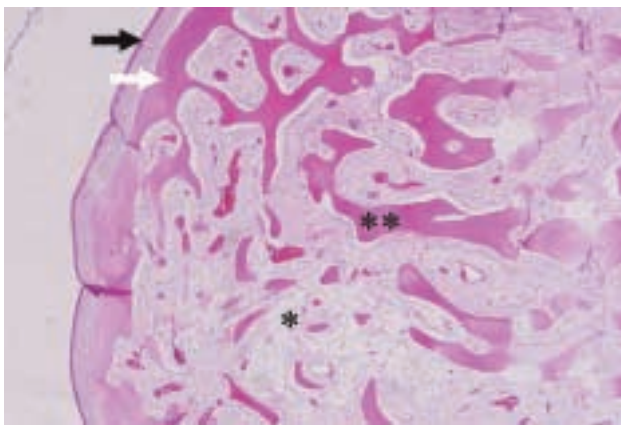

Figure 4. Histopathology of the osteolipoma (HE, X40). Black arrow: external ear canal skin, white arrow: dense osseous cortical bone cover, *: mature adipose and fibrous tissue, **: bony trabeculae showing focal osteoblastic and osteoclastic activity.

produce symptoms by compression. Depending on the location, symptoms and signs such as the sensation of a lump, discomfort during chewing and swallowing, facial asymmetry, tinnitus, blocked ear and reduced hearing can be present [3,7,8,16,17,19,22]. In our case, the patient had slight fullness and moderate conductive hearing loss on left ear which might be due to blocked ear canal by osteolipoma or impacted wax in medial part of the ear canal.

The CT scan is extremely useful in evaluation of the osteolipoma. CT scan is of great value for diagnosis by revealing the fatty density such as hypodens areas of the mass and the presence of the osseous elements with surrounding hyperdens areas or calcification. CT may also assess the exact site and extent of the growth [16,17,20]. In the present case, CT images revealed a well demarcated mass including bony density elements within, and covered by an osseous shell in the entrance of the left ear canal.

Lipomas show signal intensity similar to that of both subcutaneous and bone marrow fat on all sequences on MRI. It is important to obtain images with a fat-suppression technique to differentiate fat from other tissues that may have similar appearance. Calcifications, compact bone and fibrous connective tissue appear hypointense areas on MRI [17,23]. In the case reported herein, MRI demonstrated approximately 1 cm mass which had heterogenic signal intensity on Tl and T2-weighted images and suppressed signal intensity on fat-suppressed images. On MRI the dense osseous cortical bone cover that was seen on CT showed a hypointense cortical line circumscribing a fatty core.

Histogenesis of the osteolipoma is still unclear. The tumor may originate from multipotent mesenchymal cells or alternatively may be metaplasia from pre-existing lipoma cells [11]. Some researchers consider such types of lipoma as benign mesenchymomas, because both adipose and chondroid or osseous components originate from mesenchymal tissues. The hypothesis is that the neoplastic transformation occurs in a mixture of several types that later differentiate into lipoblasts, chondroblasts or osteoblasts and fibroblasts. Another hypothesis is that only the adipose cells undergo a neoplastic transformation, and the cartilage and bone are produced by metaplasia of fibroblasts to chondroblasts or osteoblasts. The structure and function of some fully modulated mesenchymal cells can change with altered systemic or local influences such as metabolic, mechanical (trauma, friction) and/or circulatory factors. Prolonged ischemia may lead to infarction, hemorrhage and calcification [7,24,25]. Although osteolipomas in the head and neck region are relatively smaller in size, cartilaginous or osseous metaplasia is mainly encountered in lipomas of large size and of long duration. Mostly, osteolipomas have been reported in middle-aged or elderly patients with a very long history measured in years [3,7,19,22]. Two cases of congenital osteolipoma which had been growing slowly since after the birth have been reported [14,15] and the other cases had long existence duration in years [3,7,19,22]. The case in our report had first noted the lesion in childhood, and it had grown continually. In our opinion, it may be possible that osteolipomas are congenital lesions and being noticed lately when become symptomatic.

Differential diagnosis includes benign tumors which may contain bone, including teratomas or dermoids. Tumor calcinosis and calcification must also be considered. Other conditions, such as ossifying fibromas, myositis ossificans and osteosarcomas, have to be taken into consideration, but imaging and histology should help in the differential diagnosis, since the usually diffuse fine pattern of mature ossification and bland mature fatty tissue is unique to the ossifying lipoma [2]. The CT differential diagnosis of a calcified mass should include a calcified lipoma, ossifying fibroma, an osteoma and an enchondroma as well as chondroblastoma or osteochondroma [11]. On MRI, old hematomas may have a similar appearance with lipomas [17].

Lipomas with osseous changes have the same prognosis as plain lipomas. Surgical excision is the treatment of choice and lipomas are usually cured by simple excision [1,11]. There has been no reported case of malignant transformation of an osteolipoma. Osteolipomas are benign and do not recur but, resection can be difficult because of their tendency to adhere to the neighboring tissues [22]. In the case reported herein the lesion was

completely excised by blunt dissection and there was no evidence of tumor recurrence in one year follow-up.

In conclusion, osteolipomas are rare but should be considered in the differential diagnosis of well demarcated masses located in the external ear canal which have suspicious physical examination findings and characteristic radiological appearance. Osteolipomas have clear histological findings and can be treated and usually cured by simple excision.

References

1. Rosenberg AE. Soft Tissue Tumors and Tumor-Like Lesions. In: Kumar V, Abbas AK, Fausto N, editors. Kumar: Robbins and Cotran: Pathologic Basis of Disease. China: Elsevıer Saunders; 2005. p. 1316-24

2. Obermann EC, Bele S, Brawanski A, Knuechel R, Hofstaedter F. Ossifying lipoma. Virchows Arch 1999; 434:181-3

3. Kameyama K, Akasaka Y, Miyazaki H, Hata J. Ossifying lipoma independent of bone tissue. ORL J Otorhinolaryngol Relat Spec 2000; 62:170-2

4. Godby AF, Drez PB, Fıeld JL. Sublingual lipoma with ectopic bone formation. Report of a case. Oral Surg Oral Med Oral Pathol 1961; 14:625-9.

5. Hughes CL. Intraoral lipoma with osseous metaplasia. Report of a case. Oral Surg Oral Med Oral Pathol 1966; 21:576-8.

6. Allard RH, Blok P, van der Kwast WA, van der Waal I. Oral lipomas with osseous and chondrous metaplasia: report of two cases. J Oral Pathol 1982; 11:18-25

7. Piattelli A, Fioroni M, Iezzi G, Rubini C. Osteolipoma of the tongue. Oral Oncol 2001; 37:468-70

8. Castilho RM, Squarize CH, Nunes FD, Pinto Júnior DS. Osteolipoma: a rare lesion in the oral cavity. Br J Oral Maxillofac Surg 2004; 42:363-4

9. Saghafi S, Mellati E, Sohrabi M, Raahpeyma A, Salehinejad J, Zare-Mahmoodabadi R. Osteolipoma of the oral and pharyngeal region: report of a case and review of the literature. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 2008;105:30-4.

10. Schneider J, Swoboda R. Oropharyngeal lipoma with osseous metaplasia. Zentralbl Allg Pathol 1987; 133:249-51.

11. Durmaz A, Tosun F, Kurt B, Gerek M, Birkent H. Osteolipoma of the nasopharynx. J Craniofac Surg 2007; 18:1176-9

12. Abdalla WM, da Motta AC, Lin SY, McCarthy EF, Zinreich SJ. Intraosseous lipoma of the left frontoethmoidal sinuses and nasal cavity. Am J Neuroradiol 2007; 28:615-7.

13. Cil Y, Kocer NE, Yapici AK. Frontal parosteal lipoma. J Craniofac Surg 2008; 19:1122-5.

14. Shuangshoti S, Suwanwela C, Suwanwela N. Congenital osteolipoma of the skull. Arch Otolaryngol 1982; 108:454-7

15. Marchac D, Arnaud E. Massive osteolipoma of the skull. Br J Plast Surg 2003; 56:177-80

16. Hazarika P, Pujary K, Kundaje HG, Rao PL. Osteolipoma of the skull base. J Laryngol Otol 2001; 115:136-9

17. Minutoli F, Mazziotti S, Gaeta M, Vinci S, Mastroeni M, Blandino A. Ossifying lipoma of the parapharyngeal space: CT and MRI findings. Eur Radiol 2001; 11:1818-21

18. Shetty D, Lakhkar B. Osteolipoma. Indian J Radiol Imaging 2002; 12:79-80.

19. Ohno Y, Muraoka M, Ohashi Y, Nakai Y, Wakasa K. Osteolipoma in the parapharyngeal space. Eur Arch Otorhinolaryngol 1998; 255:315-7

20. Blanshard JD, Veitch D. Ossifying lipoma. J Laryngol Otol 1989; 103:429-31.

21. Dutescu N, Georgescu L, Hary M. Lipoma of submandibular space with osseous metaplasia. Report of a case. Oral Surg Oral Med Oral Pathol 1973; 35:611-5.

22. Turkoz HK, Varnali Y, Comunoglu C. A case of osteolipoma of the head and neck area. Kulak Burun Bogaz Ihtis Derg 2004;13:84-6

23. Kawashima A, Magid D, Fishman EK, Hruban RH, Ney DR. Parosteal ossifying lipoma: CT and MR findings. J Comput Assist Tomogr 1993; 17:147-50

24. Enzinger F, Weiss S: Benign lipomatous tumours. In Soft tissue tumors. St Louis: Mosby; 2001. p. 574-81

25. Fujimura N, Enomoto S. Lipoma of the tongue with cartilaginous change: a case report and review of the literature. J Oral Maxillofac Surg 1992; 1015-7