Osteolipoma Presenting as a Submandibular Mass: A Rare Presentation

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Abstract Osteolipomas are rare tumors, particularly in the head and neck region, in contrast to lipomas which are the most common benign neoplasms in this location. Osseous changes are benign and mainly occur in long-standing lipomas. Despite its rarity, this neoplasm has a characteristic histopathologic appearance with lamellar bony spicules within the mature adipose tissue. Of the 19 cases previously reported in the oropharyngeal area, only two were located in the submandibular region. We report the third case of submandibular osteolipoma, presenting as an asymptomatic mass in a middle-aged man. Our review of head and neck osteolipomas would be of particular interest to pathologists and head and neck surgeons, in order to avoid inappropriate treatment of an otherwise benign lesion.

Keywords Osteolipoma · Submandibular · Ossifying lipoma

Introduction

Lipomas are the most common benign soft tissue mesenchymal neoplasm in adults [1] that can arise in any location in which fat is normally present. The majority occur in the upper half of the body, particularly in the trunk and the neck [2]. Lipoma can harbor elements other than fat like angioliipoma, myoliipoma, fibrolipoma, and osteoliipoma.

Osteoliipoma more commonly occurs in older individuals with a long history of persistence and slow progression [3]. It is a rare tumor that has been reported in various anatomical locations, being even more rare within the oral and pharyngeal region. A total of 19 cases of oropharyngeal osteoliipomas have been reported (Table 1) [3–10]. Ten patients, who had intraoral masses in the buccal mucosa, alveolar mucosa, palate, or tongue, were either asymptomatic or presented with facial swelling. Another major presentation was a parapharyngeal or nasopharyngeal mass causing dysphagia, hearing loss, jaw pain or paresthesia.

Only four cases of this rare type of lipoma presented as neck masses [4, 11], two of which reported in 1973 and 1998 in the submandibular area [12, 13]. We are reporting the third case in this special location.

Case Report

A 67-year-old male was referred to the otolaryngology clinic for evaluation of an asymptomatic mass in the left submandibular area (Fig. 1). The mass had been slowly

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Table 1 Osteolipomas reported in the oral, parapharyngeal, and neck regions

Case report	Location	Sex/age	Clinical presentation
Godby et al. [14]	Oral, floor of the mouth	M/54	
Hughes [15]	Oral, right mandibular buccal vestibule	M/69	Facial asymmetry
Allard et al. [16]	Oral, left mandibular buccal vestibule	F/81	Facial asymmetry
Piattelli et al. [17]	Oral, tongue	F/49	Asymptomatic
Minutoli et al. [18]	Right parapharyngeal space	F/46	Dysphagia, hypoacusia
Shetty and Lakhkar [19]	Right parapharyngeal space	F/17	Dysphagia, hypoacusia
Castilho et al. [20]	Oral, buccal mucosa	F/65	Asymptomatic
Turkoz et al. [4]	Neck	F/34	
Durmaz et al. [21]	Nasopharynx, left soft palate defect	M/21	Hypoacusia
Saghaphi et al. [3]	Oral, right mandibular alveolar mucosa	M/68	Asymptomatic
Gokul et al. [5]	Oral, hard palate	M/6	Congenital cleft palate
Juliasse et al. [6]	Oral, buccal sulcus		
De Castro et al. [7]	Oral, buccal mucosa	F/47	Facial asymmetry
Bulkeley et al. [8]	Left parapharyngeal space	M/68	Jaw pain and numbness
Bohm et al. [9]	Left parapharyngeal space	F/15	Asymptomatic
Adebiyi et al. [10]	Oral, palate	F/37	Asymptomatic
Palade et al. [11]	Neck		
Ohno et al. [12]	<i>Right submandibular space</i>	<i>F/58</i>	<i>Submandibular swelling</i>
Dutescu et al. [13]	<i>Left submandibular space</i>	<i>M/40</i>	<i>Submandibular swelling</i>

The two cases in italics indicate the only cases previously reported in the submandibular region

Fig. 1 Clinical photograph of the left submandibular mass

growing for the past 10 years without causing pain, paresthesia, or dysphagia. The systemic review was not contributory. His past medical history was significant for a mass in his left arm which was previously excised and diagnosed as lipoma. A head and neck evaluation revealed a firm, non-tender, and mobile mass in the left submandibular area, approximately 4 cm in its largest diameter, with no changes in the overlying skin. The neck was supple with no palpable lymphadenopathy. The thyroid gland was non-palpable. No bruits, thrills, or venous distention were detected. Endoscopy showed a normal pharynx and larynx. Laboratory studies, including a total serum calcium level, were unremarkable.

Contrast-enhanced Computerized Tomography scan of the neck demonstrated a 3.5 × 2.2 cm well-circumscribed

mass within the left submandibular space anterior to the submandibular gland. This mass had a fatty capsule and a central densely calcified portion, measuring 2.6 cm in its largest diameter. A mild mass effect was present on the anterior aspect of the submandibular gland (Fig. 2). Nasopharynx, oral cavity, hypopharynx, and larynx were normal. No abnormal lymphadenopathy or enhancement was seen. There were no previous images for comparison.

Excision of the left upper neck mass, located at level 1B, was performed. The mass was completely separate from the left submandibular gland. Grossly, the specimen consisted of a partially hard and partially firm yellow-tan mass measuring 3.5 × 3.0 × 2.0 cm with an outer smooth surface. On sectioning, the mass was heterogeneous with areas of pink-tan-white hard to firm tissue intermixed with the yellow adipose tissue (Fig. 3). On microscopic examination, mature adipose tissue made up the majority of the specimen. The firm areas showed bundles of collagen between mature adipocytes (Fig. 4). A shell of lamellar bone encased the hard areas with bony spicules noted within mature adipose tissue (Fig. 5).

Discussion

The large majority of soft tissue tumors are benign and at least one third of the benign tumors are lipomas. Lipoma can arise within subcutaneous tissue, within deep soft tissues, on the surfaces of bone, or between skeletal muscle fibers [1]. Osteolipomas, lipomas containing mature

Fig. 2 CT images demonstrate a well-defined encapsulated mass just anterior to the left submandibular gland

Fig. 3 Heterogeneous mass with areas of tan-white hard tissue intermixed with adipose tissue

lamellar bone irregularly distributed in the predominant adipose component, are extremely rare in clinical practice [3, 12]. Ossifying lipoma, in contrast, mainly describes an intraosseous lipoma that arises within the medullary cavity and rarely in the cortex of a bone. This term has interchangeably been used in the literature to describe osteolipoma.

The subtype of osteolipoma that is completely independent from bone is rarely reported and occurs most

Fig. 4 Fibrolipomatous area (hematoxylin–eosin, $\times 100$)

commonly in the head and neck region [1] with 19 cases being reported in the oropharynx and the neck to the best of our knowledge (Table 1). More than half of the cases were intraoral lesions [3, 5–7, 10] and a quarter were reported in the naso/parapharyngeal area [3, 8, 9]. Only four cases of osteolipoma have presented as neck masses [4, 11], two being located in the submandibular region [12, 13]. Considering the rarity of this case in the submandibular region, the purpose of this paper is to describe the third case of submandibular osteolipoma, which would be of particular interest to head and neck surgeons and pathologists [4, 11–13].

Based on data presented in Table 1, there is no clear sex preponderance in patients with oropharyngeal osteolipoma.

Fig. 5 Osteolipoma area (hematoxylin–eosin, $\times 40$)

Around 75 % of these cases were reported in patients over the age of 30, which is compatible with this case which presented in the sixth decade. Most of the reported cases had a long history of slow progression [3], and our patient had the lesion for at least a decade before seeking medical advice, emphasizing its benign natural course.

Proposed etiologies for the genesis of osteolipomas include metaplasia of fibroblasts from within the lipoma, origination of the adipocytes and osseous components from a multipotent stem cell, repetitive external microtrauma, and ischemia secondary to outgrowth of the tumor's blood supply [3, 8, 10]. Ohno has described the fibrous tissue to be the origin of osseous changes in lipomas [12]. During the surgery, this mass was readily dissected from the submandibular gland and was not attached to the mandibular bone, excluding the possibility of an ossifying lipoma. The presence of dense collagen fibers and osseous trabeculae scattered among adipocytes lend some support to the hypothesis of fibroblastic metaplasia or origination of tumor from a multipotential stem cell.

Despite the rarity of this neoplasm and its tendency to develop in different anatomic locations within the oropharyngeal region, the histopathologic appearance of lamellar bony spicules within the adipose tissue is quite characteristic. In conclusion, osteolipoma should be considered in the differential diagnosis of benign neoplasms in this location.

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