Radiology Case Reports 13 (2018) 361–364

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journal homepage: http://Elsevier.com/locate/radcr

Head and Neck

Large intramuscular lipoma of the tongue

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ARTICLE INFO

Article history:
Received 2 December 2017
Accepted 4 January 2018
Available online 3 February 2018

Keywords:
Oral lipoma
Tongue
Intramuscular lipoma
Infiltrating lipoma

ABSTRACT

We describe a case of a 57-year-old man referred to an oral maxillofacial surgeon for a nontender, large intramuscular tongue mass. A computed tomography scan with contrast showed a homogenous right tongue intramuscular fatty mass measuring 3.8 cm × 2.8 cm in the axial dimension and 2.2 cm in the craniocaudal dimension. Histologic examination revealed multiple lobulated sections of mature adipocytes and occasional entrapped skeletal muscle fibers. The final pathologic diagnosis was intramuscular lipoma. Although lipomas account for approximately 50% of all soft tissue neoplasms, intramuscular (infiltrating) lipoma of the tongue is exceedingly rare.

Introduction

Lipoma is a common benign mesenchymal tumor, accounting for 50% of all soft tissue neoplasms [1,2]. It exhibits lobular growth of mature adipocytes with demarcated borders and is often enclosed within a fibrous capsule [2]. Conventional lipoma arises most commonly in subcutaneous tissue; however, a deep-seated subtype may develop within skeletal muscle, where it is termed an intramuscular (infiltrating) lipoma. The latter subtype is relatively uncommon, accounting for less than 1% of all lipomas [3]. Histologically, intramuscular lipoma displays mature adipocytes that often irregularly infiltrate and entrap muscle fibers. The muscle fibers may show evidence of atrophy and in many places the adipocytes completely exclude muscle fibers [1,4]. Tumor vasculature consists mainly of capillaries and is often inconspicuous because of compression by surrounding adipocytes [4–6].

Oral lipomas in general are uncommon, representing approximately 1%-5% of all benign neoplasms of the oral cavity [7–12]. Intramuscular lipoma accounts for just 3%-7% of these cases, making intramuscular lipoma in this region exceedingly rare [12,13]. Here, we describe an intramuscular lipoma of the tongue, initially identified and defined through computed tomography (CT) scan.

Competing Interests: The authors have declared that no competing interests exist.

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https://doi.org/10.1016/j.radcr.2018.01.014

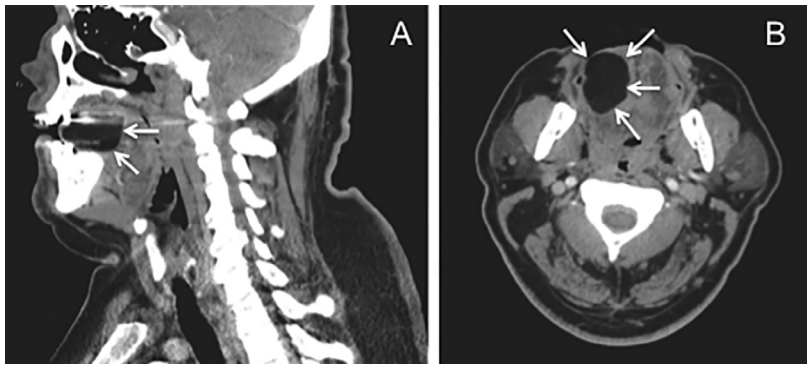

Fig. 1 – CT images of a 57-year-old male. (A) Sagittal and (B) axial scans demonstrating the large, low attenuation lipomatous mass (arrows) within the tongue. CT, computed tomography.

Case report

A 57-year-old Caucasian male was referred to an oral maxillofacial surgeon by his primary care physician because of a nontender mass in the right lateral side of his tongue. The patient had not previously noticed the mass and did not complain of any functional impairment. Physical examination by the oral surgeon revealed tongue swelling, but other aspects of the examination were unremarkable. The patient's personal and family history were also unremarkable. The preliminary differential diagnosis was determined to be angioedema of the tongue, glandular cell tumor, or dermoid cyst. The surgeon ordered a CT scan with contrast of the tongue, head, and mandible, which was performed at a local hospital, where we encountered the patient in the radiology department.

A CT scan showed a homogeneous right tongue intramuscular fatty mass, measuring 3.8 cm × 2.8 cm in the axial dimension and 2.2 cm in the craniocaudal dimension (Fig. 1). The average density of the mass was –100 Hounsfield units (HU). The impression of the CT was an intramuscular lipoma of the tongue.

The patient had the mass surgically resected and tissue was sent for pathology analysis. The resected ovoid fatty tissue fragment was serially sectioned; it revealed pale yellow, homogeneous, smooth surface sections. Measurements were 4.4 cm × 3.2 cm × 2.2 cm. There was no hemorrhage or areas of necrosis. Microscopic examination revealed multiple lobulated sections of mature, univaculated adipocytes of relatively uniform size and shape with occasional entrapped skeletal muscle fibers. There was no evidence of hyperchromasia, pleomorphism, or multinucleation of adipocytes and no evidence of lipoblasts (Fig. 2). The histopathology was consistent with the final diagnosis of intramuscular lipoma.

The postoperative period was uneventful. Verbal follow-up was conducted at 10 months and the patient reported no signs or symptoms of recurrence. The patient's tongue function and appearance are normal. Although there was no overt evidence of lipoblastic atypia at the time of the surgical resection, this lesion may locally recur.

Discussion

The patient presented in this report is a 57-year-old male who was diagnosed with an intramuscular lipoma in the tongue. Intramuscular lipoma is an uncommon subtype of lipoma that typically occurs in the large muscles of the trunk and extremities [1,14,15]. Intramuscular lipoma in the tongue is particularly rare [16–18], making this case noteworthy.

The etiology of intramuscular lipoma remains unclear. McTighe and Chernev [4] point to similarities with other lipoma subtypes and suggest intramuscular lipoma likely represents a true neoplasm directly originating from multipotent mesenchymal cells. Others have suggested trauma, chronic

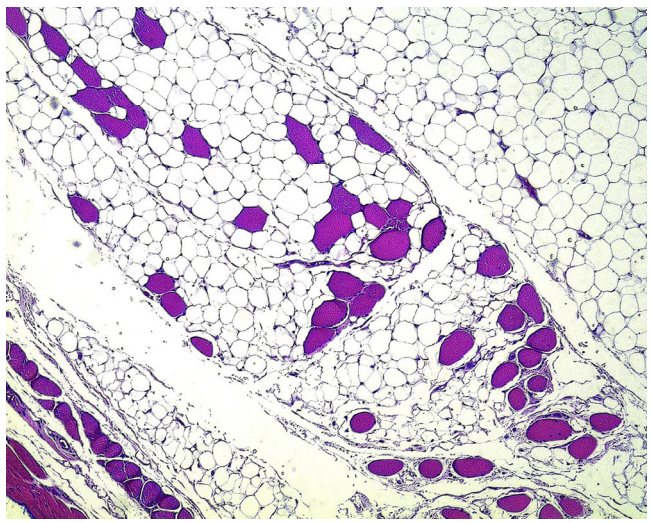

Fig. 2 – Histopathology of the intramuscular lipoma (hematoxylin-eosin). Adipocytes are seen infiltrating the skeletal muscle of the tongue and entrapping muscle fibers. The tumor displays mature, univaculated adipocytes of fairly uniform size and shape. No lipoblasts are seen. Original magnification 100×.

irritation, obesity, hormonal imbalance, and metabolic conditions as possible causative agents [19,20]. Unlike subcutaneous lipomas, where multiple lipomas are familial in approximately 30% of cases [2], there appear to be no reports of familial cases of intramuscular lipomas [4] and there is a lack of evidence to suggest any type of genetic transmission. This observation falls in line with the present case, in which the patient reported no known family history of intramuscular lipoma. Interestingly, Mori et al. [21] provide evidence that atrophy or degeneration of select muscle fiber types promotes infiltrative growth of intramuscular lipoma, suggesting an association with focally neurogenic or myogenic disorders in the lesion.

For subcutaneous lipomas, most pose no diagnostic dilemmas and a presumptive diagnosis may be made clinically. In contrast, intramuscular lipomas typically require radiographic consultation. CT scan and magnetic resonance imaging are the choice modalities for identifying adipose tissue and for defining the size, depth, and area of the mass along with its anatomic relationship with surrounding structures. By CT scan, the low mass density of the lipoma, with HU values in the negative range, is used to confirm the presence of adipose tissue. In the case presented here, the attenuation range was –100 HU, which falls within the normal values for adipose tissue [22]. For intramuscular lipomas with an infiltrative nature, CT scan often reveals the entrapment of muscle fibers. These appear as soft tissue density streaks of variable thickness and occasional interruption [2,23].

In many cases, the imaging characteristics of intramuscular lipoma and well-differentiated liposarcoma closely resemble one another [2,4,23–25] and therefore radiological imaging alone is insufficient for a diagnosis. It is essential that histopathologic examination be included in establishing the definitive diagnosis. A distinguishing feature between intramuscular lipoma and liposarcoma is the presence or absence of lipoblasts. This cell stage is absent in lipoma but is recognized as a hallmark of liposarcoma. Additionally, lipoma consists of mature adipocytes of uniform size and possesses minimal vascularization, whereas liposarcoma displays cellular pleomorphism, hyperchromasia, and marked vascularization [1,4,15].

Lipomas, including intramuscular lipomas, generally are not harmful; however, in the oral cavity the mass or bulk effect can obstruct mastication or impede speech [16–18]. The definitive treatment is surgical resection. Unlike other lipoma subtypes, intramuscular lipoma has a reported high rate of recurrence. Recurrence rates have been reported between approximately 50% and 80% [19,24,26] and recurrence can occur years after excision [27]. These high rates can be attributed to the infiltrating tendencies of intramuscular lipoma and the resulting difficulty removing the entirety of the mass [4,15,17]. Han et al. [15] report success in dramatically reducing the recurrence rates of intramuscular lipoma by using a wide excision margin, as would be the case during the resection of a malignant tumor. In the present case, the patient reported no signs or symptoms of recurrence 10 months postoperative; however, given the infiltrative nature of his tumor demonstrated by histopathology, the lesion may locally recur.

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