Osteochondrolipoma of the Submandibular Region: A Case Report and Review of the Literature

Raoulin Soulard · Anh Tuan Nguyen ·
Jean-Baptiste Souraud · Pierre-Antoine Oddon ·
Bernard Fouet · Olivier Cathelinaud

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Abstract Lipoma is an extremely common and ubiquitous benign soft tissue tumor composed of mature adipose tissue, with frequent differentiation towards other mesenchymal elements such as blood vessels, fibrous tissue, or muscle. They are typically slow-growing, encapsulated, superficial or deep, and have a wide range in size depending on their location. The presence of cartilage or bone is very rare, with most reported cases in the head and neck area. We report the case of an osteochondrolipoma of the submandibular region with associated benign bone marrow elements, with a review of the literature and discussion of the differential diagnosis.

Keywords Lipoma · Osteolipoma · Ossifying lipoma · Parosteal ossifying lipoma · Osteochondrolipoma

Introduction

Lipoma is the most common soft tissue tumor and causes few clinical problems. Most present during middle age in superficial (subcutaneous) locations as the upper back, shoulder, abdomen and proximal extremities. It has been estimated that approximately 13 % of lipomas occur in the head and neck [1]. Lipomas and lipoma variants occur infrequently in the oral and maxillofacial region, most commonly in adult males in the parotid region, followed

closely by the buccal mucosa, the margin of the tongue and the lower lip [2–4]. Some are deep-seated (subfascial) and may be more diagnostically challenging, both radiologically and histologically. Admixed differentiation into a diversity of mesenchymal elements, such as fibrous tissue (fibrolipoma), blood vessels (angiolipoma) or muscle (myolipoma) is a relatively frequent event. Osseous change is extremely rare, mainly in large and long-standing lipomas [5, 6]. We report our experience in managing a patient with a submandibular mass with the configuration of a lipoma and with concurrent bone and cartilaginous differentiation, consistent with an osteochondrolipoma.

Case Report

A 61-year-old man presented with a slow-growing painless mass of the right submandibular region, with recent discomfort. He remembered that a surgeon, more than 20 years ago, had advised him to have the lesion resected. The patient had type 2 diabetes and no other significant past medical history. He could not formally exclude a prior minor trauma to the region. On exam, there was a single well-defined mobile soft tissue mass, with normal surrounding skin, and no lymphadenopathy. Intraoral examination showed an intact mucosa.

CT revealed an oval-shaped smooth mass of fat density measuring $4.5 \times 4.5 \times 4.4$ cm with an area of prominent ossification measuring $3.7 \times 3.2 \times 2.7$ cm without connection to the mandible (Fig. 1a, b).

Under general anesthesia, the tumor was readily dissected manually, but was focally adherent to the underlying periosteum, and thus needed a subperiosteal dissection.

Gross examination showed a yellowish mass of 26 g measuring $4.5 \times 4.5 \times 4$ cm with a smooth surface. It was encapsulated by a thin fibrous capsule. After serial sectioning,

R. Soulard (✉) · A. T. Nguyen · J.-B. Souraud · B. Fouet
Department of Pathology, Sainte-Anne Naval Hospital,
BP20545, 83041 Toulon Cedex 9, France
e-mail: soulard65@gmail.com

P.-A. Oddon · O. Cathelinaud
Department of Otolaryngology, Sainte-Anne Naval Hospital,
BP20545, 83041 Toulon Cedex 9, France

Fig. 1 a–b CT scan shows a right submandibular *oval-shaped* smooth mass of mixed density, with fat attenuation and areas of ossification, and neither contact with the mandible nor underlying

bone reaction. **c** Multilobular, finely encapsulated *yellowish* mass. **d** Cut surface shows *yellow-tan* glistening fat intermixed with bone and solid whitish areas

adipose tissue with fibrous and bony streaks were visible, as well as some whitish-translucent areas (Fig. 1c, d).

After decalcification, histologic examination revealed abundant benign-appearing adipose tissue with haphazardly distributed islands or lamellar structures of mature bone including osteocytes. Woven bone with rims of osteoblasts and osteoclasts were occasionally seen, as well as patchy cartilaginous areas, sometimes associated with endochondral ossification. Many of these bone spicules were of desmal-type ossification (typically seen in heterotopic bone formation [7]) surrounded on one of their surfaces by periostum-like plaques of fibro-myxoid mesenchymal tissue. No lipoblasts and no mitoses were seen. Trilineage hematopoietic cell islands were noted (Fig. 2, 3, 4, 5). A diagnosis of lipoma with bone and cartilaginous differentiation consistent with osteochondrolipoma was rendered.

Discussion

Osseous differentiation in lipoma is a rare event. In a series of 635 lipomas seen over a 5-year period, only 6 cases with

Fig. 2 Fatty tumor with a focus of lamellar bone with attached periosteum-like mesenchymal tissue and central bone marrow elements, giving a lagoon-like aspect

ossification were found [8]. Authors have been using many terms to describe lipomas with osseous differentiation: osteolipoma, osseous lipoma, ossified lipoma, ossifying lipoma, lipoma with osseous metaplasia, lipoma with

Fig. 3 Area of osseous and cartilaginous differentiation

Fig. 5 Trilineage mature hematopoiesis

Fig. 4 **a** Fibro-myxoid mesenchymal bud. **b** Early osteogenesis. **c** Woven bone with intense osteoblastic activity. **d** Osteo-cartilaginous differentiation

osseous change, ossifying parosteal lipoma, parosteal ossifying lipoma, parosteal lipoma with osseous differentiation, lipomatous tumor with osseous differentiation, and benign mesenchymoma. According to the World Health Organization (WHO) 2002 classification of soft tissue tumors, conventional lipoma is composed of lobules of mature adipocytes and can arise within subcutaneous tissue

(superficial lipoma) or within deep soft tissues (deep lipoma) or even on the surfaces of bone (parosteal lipoma) [9]. Intraosseous lipomas arise within bone. Currently, the terms *osteolipoma* and *ossifying lipoma* are generally used synonymously for soft tissue tumors without relationship to the underlying bone, while *ossifying parosteal lipoma* is used if the tumor is intimately associated with the

Table 1 Previously reported cases of lipomas with osseous differentiation in the head and neck (extra cranial-extraspinal)

NO.	Author	Age	Sex	Location	Size (cm)	Duration	Diagnosis
1	Godby [24]	54	M	Sublingual	7 × 6 × 3	1 year	OL
2	Hughes [25]	69	M	Right mandibular vestibule	3.5 × 2.6 × 1.7	–	OL
3	Dutescu [26]	40	M	Left submandibular space	7 × 4	3 years	OL
4	Allard [27]	81	F	Left mandibular vestibule	3.5 × 2	>30 years	OL
5	Blanshard [28]	–	–	Retropharyngeal	–	–	OL
6	Ohno [5]	58	F	Right parapharyngeal/mandible	9 × 4	1 year	OL
7	Kameyama [29]	46	M	Neck	3 × 2	–	OL
8	Hazarika [30]	17	F	Right parapharyngeal space	5 × 4	1 year	OL
9	Piatelli [31]	49	F	Tongue (left lateral margin)	0.8	8 years	OL
10	Minutoli [32]	46	F	Right parapharyngeal space	2.5 × 4	–	OL
11	Shetty [33]	17	F	Right parapharyngeal space	–	1 year	OL
12	Castilho [6]	65	F	Left buccal mucosa	1 × 1 × 0.8	–	OL
13	Durmaz [34]	21	M	Nasal septum/nasopharynx	1.3 × 1.9 × 2.6	5 years	OL
14	Saghafi [35]	68	M	Mandibular mucosa	1.8 × 1.2 × 1	4 years	OL
15	Kumar [36]	50	F	Right upper eyelid	2 × 1.5 × 1	5 years	OL
16	Kuyama [4]	59	M	Left lower labial vestibule	0.9 × 0.5 × 0.5	2 months	FL/OL ^a
17	Juliasse [3]	–	–	Buccal sulcus	–	–	OL
18	De Castro [37]	47	F	Left buccal mucosa	1.5	–	OL
19	Diom [38]	21	F	Left parotid/parapharyngeal	5	1 year	OL
20	Adebiyi [39]	37	F	Left side of the palate	3.2 × 3 × 1.6	10 years	OL
21	Bohm [40]	15	F	Left parapharyngeal mass	3.8 × 3.5 × 2.8	–	OL
22	Hsu [41]	71	M	Left buccal mucosa	3.8 × 2.4 × 1.3	4 years	OL
23	Bulkeley [42]	68	M	Left parapharyngeal space	4 × 1.5 × 1	–	OL
24	Sun [43]	48	M	Mandible	7 × 5 × 5	>20 years	OPL
25	This study (2012)	61	M	Submandibular region	4.5 × 4.5 × 4	>20 years	OCL ^{a,b}

OL osteolipoma, FL fibrolipoma, OPL ossifying parosteal lipoma, OCL osteochondrolipoma

^a With chondroid differentiation

^b With mature hematopoiesis

periosteum. A review of the English literature found that, outside the head and neck area, lipomas with osseous differentiation have been reported in deep locations adjacent to bone tissue in the thigh [10–13], knee [14, 15], sterno-clavicular region [16], shoulder [17], wrist [14], hand [18], intraspinal [19] or in superficial subcutaneous tissue [20, 21]. More than 60 % of the cases occur in the head and neck region, where they have been described in intraspinal [22], intracranial [23] (suprasellar/hypothalamic and interhemispheric) and extracranial-extraspinal locations [3–6, 24–42]. The latter are summarized in Table 1. There is no clear sex predominance (12 women, 11 men, 2 unknown) and the average patient age is 48.2 years (SD, 19.4; range 17–81 years). The clinical history is usually that of a painless, slow-growing mass often measured in years (mean more than 7 years, ranging from 2 months to more than 30 years). Average tumor size is 3.8 cm (SD, 2.2 cm; range 0.8–9 cm).

Osteolipomas are well-encapsulated and usually have a sharp transition between bone and adipose tissue. However

there are 3 reports of osteolipomas with bony attachments [24, 30, 34]. In these cases, the adipose component was predominant and the mature lamellar bone tissue was irregularly distributed, with a periosteum-like fibrous or fibro-myxoid tissue adherent to one of the surfaces of the bone spicules. Cartilaginous differentiation may be seen. In the head and neck, only 2 cases, including our report had both bony and cartilaginous differentiation [4]. Excluding our case, hematopoiesis has only been reported twice in an osteolipoma [20], never in a head and neck location. Although our tumor would fit the description of osteolipoma, its attachment to the periosteum, however, would be in favor of an ossifying parosteal lipoma. Only 9 cases of ossifying parosteal lipomas have been described in the literature: 7 in the upper extremities (femur 5, humerus 2), 1 in the trunk (rib) and 1 in the mandible [43–51]. An underlying bone reaction was always present (uneven and undulating cortex, hyperostosis, osseous excrescences), but was absent in our report. The pathogenesis of these lesions might give us the answer. Two main theories exist. These

tumors may originate directly from a multidirectional differentiation capacity of adipose tissue-derived stem cells [10, 14, 20], or they may arise from secondary metaplasia of fibroblasts after repetitive trauma, metabolic changes, or ischemia [14, 52]. The few cytogenetic studies on osteolipomas and parosteal lipomas show they share the same t(3;12) translocations as conventional soft tissue lipomas [14, 15, 53, 54]. Matsui et al. detected an identical HMGA2-LPP fusion transcript in both the fibro-adipose and the chondro-osseous components of a tendon lipoma, suggesting that the HMGA2-LPP fusion protein induces fibroadipogenesis as well as osteochondrogenesis [15]. In our case, routine hydrochloric acid decalcification resulted in failure to obtain DNA for in situ hybridization. Decalcification using an EDTA solution allows for the performance of interphase FISH for HMGA2 in 4–5 µm paraffin-embedded thin sections using bacterial artificial clones flanking the HMGA2 locus on 12q15. These studies provide evidence that both these tumors are variants of conventional lipoma. In our case, the diagnosis would probably have been consistent either with an osteolipoma or an ossifying parosteal lipoma. However, as foci of cartilaginous differentiation were observed, we favored an osteochondrolipoma. This attractive term was coined by Rau et al. [7] to describe an ossifying parosteal lipoma of the thigh with cartilaginous differentiation, and used by Gru et al. [20] to describe a subcutaneous osteolipoma with chondroid differentiation. It accurately describes the composition of the tumor, independently of its localization, and remains a benign adipocytic neoplasm that usually does not recur after complete excision.

The differential diagnosis of osteochondrolipoma is limited. Mesenchymal hamartomas nearly always present in neonates or childhood and are uncommon in the head and neck, where they have been described in the sinonasal tract, nasal cavity, nasopharynx, oropharynx, hypopharynx, larynx, trachea, cervical esophagus, ear and eustachian tube. They can be very large tumors and partially cystic. Microscopically, the hamartomatous nature is confirmed by the disorganized admixture of mature elements: fibrous tissue, fat, vascular spaces, smooth muscle, nerves, cartilage, and bone trabeculae that commonly contain hematopoietic marrow. Osteoclast-like giant cells, aneurysmal bone cyst-like areas, hemosiderin and foam cells may be so extensive as to obscure the underlying hamartomatous elements, and focal areas of stromal hypercellularity can sometimes raise concern for a sarcoma. In the head and neck, a majority of hamartomas are composed of vascular tissue (angiomas), and only one hamartoma with bone formation has been reported [55]. Other tumors including teratoma, ossifying fibroma, myositis ossificans, and extraskelatal osteochondroma should also be discussed; however, they do not have adipose tissue as their main

component. Sarcomas can show calcification or ossification including liposarcoma, synovial sarcoma, extraskelatal osteosarcoma, and extraskelatal chondrosarcoma. Overdiagnoses of well-differentiated liposarcoma can occur due to the presence of non-lipomatous areas within lipomas [56]. Clinicians should be aware that clinical and radiological features with head and neck osteolipomas may mimic malignant neoplasms. Bulkeley et al. described an osteolipoma of the parapharyngeal space with jaw pain and numbness in the maxillary (V-1) distribution with a presumptive diagnosis of liposarcoma after CT angiography [42]. Nevertheless, the overall dimensions of the lesion were less than 10 cm, and the tumor was greater than 75 % fat radiologically, both favoring a benign diagnosis. Histology helps in the differential diagnosis, since the usually diffuse fine pattern of mature ossification and bland mature adipose tissue is unique to the ossifying lipoma.

In summary, osteolipomas or ossifying lipomas are rare variants of lipomas that are increasingly recognized in the head and neck region. When cartilaginous differentiation is present, a more appropriate term might be osteochondrolipoma. Our case represents the first description of an osteochondrolipoma associated with mature hematopoiesis in the head and neck area.

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