

# Osteolipoma of Parotid Gland: A Rare Presentation

**Jing Hu**

Sun Yat-Sen University

**Nan Xie**

Sun Yat-Sen University

**Zehang Zhuang**

Sun Yat-Sen University

**Cheng Wang** (✉ [wangch75@mail.sysu.edu.cn](mailto:wangch75@mail.sysu.edu.cn))

Sun Yat-Sen University <https://orcid.org/0000-0001-8155-5064>

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## Case report

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## Abstract

## Background

Osteolipoma is an uncommon benign tumor containing mature adipose tissue with bone metaplasia foci. It is rarely observed in head and neck region as compared to the lipoma, especially in the region of parotid gland.

## Case presentation:

We report the first case of intraparotid osteolipoma with its clinical, radiographic and pathological findings. A 47-year-old men presented with an immovable mass in his left cheek with a 10-year evolution. Magnetic resonance imaging (MRI) scans showed a 3.2-cm fat-containing mass involving in the left parotid. The patient was taken to the operating room for excision of the mass and the final pathologic diagnosis was osteolipoma. Due to its rarity and clinical significance, we also reviewed the osteolipoma presenting in major salivary gland regions.

## Conclusion

Intraparotid osteolipoma is an extremely rare neoplasm originating from adipogenic tissue, which should be taken into consideration in the differential diagnosis of parotid tumor.

## Background

Osteolipoma is a benign mesenchymal neoplasm originating from fatty tissue with bone formation, which is a rarer histological variant among lipomas and accounts for less than 1% of cases reported in the literature<sup>1,2</sup>. It has been documented in the upper and lower extremities, spine, subcutaneous soft tissue and head and neck region<sup>1-5</sup>. To the best of our knowledge, only one case of osteolipoma in the parotid region was documented in the English language literature, which was reported by Diom et al.<sup>6</sup> in 2011. They described a 21-year-old female patient with osteolipoma located between the deep lobe of the parotid gland externally and the parapharyngeal space internally, indicating the tumor was independent from the parotid gland and should be classified as periparotid osteolipoma. Here, we describe the first case of intraparotid osteolipoma located at the superficial lobe of parotid gland. Moreover, we also reviewed the osteolipoma presenting in major salivary gland regions due to its rarity and clinical significance.

## Case Presentation

A 47-year-old male patient was admitted to the department of Oral and Maxillofacial Surgery at Hospital of Stomatology, Guanghua School of Stomatology, Sun Yat-sen University. He presented a slowgrowing and painless mass in the left cheek with a 10-year evolution. The patient had a history of trauma in that region in 10 years ago. The medical history and systemic review were noncontributory. Physical examination showed a 2.5 × 3.2 cm<sup>2</sup>, painless, hard, barely mobile mass with a smooth surface in the left parotid region (Fig. 1A). There was no intraoral expression and peripheral facial palsy. After the physical examination, the initial impression was diagnosed as the tumor of parotid gland and MRI scanning was requested. MRI findings revealed that a 2.5 × 2.0 × 3.2 cm<sup>3</sup>, well-defined oval lipomatous mass located in the superficial lobe of left parotid with high signal intensity on T2-weighted image and suppressed signal intensity on fat-suppressed T1 and T2-weighted image (Fig. 1B-E). A hypointense line was also observed on the MRI images, indicating that the fatty mass was circumscribed with an ossifying layer (Fig. 1E). However, the internal boundary of tumor with parotid gland is not clear (Fig. 1F). Based on these clinical and radiographic findings, the differential diagnosis including pleomorphic adenoma with ossification, osteolipoma, chondrolipoma, sialolipoma and other salivary gland tumors was considered. Under general anesthesia, surgical removal of tumor was done with a standard preauricular/cervical incision. After exposure the parotid gland, the facial nerve was identified and preserved. Then, a yellowish tumor was dissected and resected completely. In agreement with the MRI findings, we found that the tumor was attached to the adjacent parotid gland at the internal boundary during the operation. After removal of the tumor, the surgical specimen was then sent to the Department of Oral Pathology, Hospital of Stomatology, Guanghua School of Stomatology, Sun Yat-sen University. Macroscopically, the resected specimen was a well circumscribed and yellowish mass with hard consistency (Fig. 2A). On sectioning, the mass demonstrated a yellow soft tissue with various intermixed ossifying structures and a fibrous capsule (Figs. 2B). Microscopic examination in paraffin embedded section revealed the tumor comprising predominantly of mature adipose tissue partly encapsulated by osseous bone shell and admixed with areas of bony trabeculae, along with intervening fibrous connective tissue, few inflammatory cells and blood vessel (Fig. 2C-F). The final established diagnosis was the 'osteolipoma'.

## Discussion

Lipomas are common benign mesenchymal neoplasm of soft tissue in various areas of body, but account for only 0.2–0.8% of major salivary gland tumors and 22.5% of all benign mesenchymal salivary neoplasms<sup>7-9</sup>. Microscopically, lipoma is composed of mature adipose tissue arranged in lobules separated by fibrous septa. However, histopathological variants of lipoma might be observed, including spindle cell lipoma, fibrolipoma, myolipoma, myxolipoma, angioliipoma, sialolipomas, chondrolipoma and osteolipoma<sup>9,10</sup>. Among them, osteolipoma is an extraordinary lipoma variant characterized by mature bone formation within the neoplastic fatty tissue. In head and neck region, more than half of the osteolipoma were reported in oral cavity and naso/parapharyngeal space<sup>11</sup>, but extremely rare in parotid gland. Although oncology of the salivary gland and parotid region is tremendously rich and varied, only seven cases of osteolipoma located at the major salivary gland regions have been reported<sup>6,11-16</sup>. Considering the rarity of this case in the salivary glands, we aim to describe

the first case of intraparotid osteolipoma and summarize the osteolipoma presenting in the region of major salivary glands, which would be of particular interest to head and neck surgeons and pathologists.

As shown in Table 1, osteolipoma can be observed in all three major salivary gland regions, regardless of the proximity to bone. Two cases were occurred in the sublingual area, four in the submandibular space and two in the parotid region. Among these cases, the submandibular osteolipomas, sublingual osteolipomas and one periparotid osteolipoma were all independent from salivary glands, indicating that the tumor might originate from fat tissue in the submandibular, sublingual and parapharyngeal space. Interestingly, the tumor was resided at the intraparotid gland in our case, which may closely mimic pleomorphic adenoma with bone formation and easily misdiagnosis as pleomorphic adenoma in clinics. In addition, the age, sex, clinical presentation, tumor size, duration of the lesion, radiographic findings and managements of these cases are also described in Table 1. In this series of cases, all cases were reported in adult patients with a relative long history of slow progression and seemed to more common in men, which is similar with salivary gland lipomas<sup>9,10</sup>.

Table 1  
Osteolipoma reported in the major salivary gland regions

Authors and year	Age	Sex	Sites of lesions	Clinical presentation	Relation with salivary gland	Duration (yrs)	Tumor size (cm)	Raidographic findings	Treatment	Pathology
Godby et al. [12].1961	54	M	Sublingual area	A soft painless mass beneath the tongue	N/A	1	7 × 6	X-ray: an osseous projection was seen in the oral floor	Tumor excision	Microscopic sections show well-formed cancellous surrounding striated muscular fibrous connective tissue, and
Dutescu et al. [13]. 1973	40	M	Submandibular region (L.)	A tumor mass in the left submandibular region	NA	3	4 × 7	N/A	Tumor excision	The tumor composed of mature adipocytes, connective tissue cells, collagen fibers and fibrous osseous metaplasia similar to normal bone tissue
Ohno et al. [14]. 1998	58	F	Submandibular region (R.)	A soft immovable mass in submandibular region	Tumor was located at extra right submandibular gland, which was displaced inferiorly	1	9 × 4	CT: low density contrast suggesting fat with bone-like tissue. MRI: Signal intensities also suggested fat and dental tissues. Bone scanning with 99 m-Tc showed accumulation in the lesion.	Tumor resection	Tumor contained fat tissue and lamella bone with a marrow-like appearance containing microvesicles erythrocytes lymphocytes inside its lumen
Diom et al. [6]. 2011	21	F	Parotid region (L.)	A sublobular, painless, hard, barely mobile mass in left cheek	The tumor was independent from the parotid located between the deep lobe of the gland externally and the parapharyngeal space internally.	1	5	NA	Tumor excision and total parotidectomy with facial nerve preservation	The tumor was made of many non-atypical adipocytes metaplastic ossification in the middle. Bone metaplasia was complete with a corticospontaneous tissue with adipose mesenchyme
Kavusi et al. [11]. 2013	67	M	Submandibular region (L.)	A firm, non-tender, and mobile mass in the left submandibular area	Tumor was located at extra gland but was involved in the anterior aspect of the left submandibular gland	10	3.5 × 2.2	CT: well-circumscribed mass with low density and a central densely calcified portion	Tumor excision	Tumor are composed of mature adipocytes and lamellar bone
Aboh et al. [15]. 2015	33	M	Submandibular region (L.)	Painless swelling in the left submandibular region	N/A	Many years	N/A	MRI shows a submandibular mass with hyper- and hypo-intensity signals on T1-weighted image.	Tumor excision without damage submandibular gland	N/A

N/A: Not available, M:Male, F:Female, L: Left, R: Right

Authors and year	Age	Sex	Sites of lesions	Clinical presentation	Relation with salivary gland	Duration (yrs)	Tumor size (cm)	Radiographic findings	Treatment	Pathology
Raghunath et al. [16]. 2015	20	F	Sublingual area	A well circumscribed, firm and painless mass	N/A	3	6 × 6	CT scanning reveals a well-defined, hypodense lesion (- 35 to - 65 HU) measuring about 6 × 6 cm within which irregular hyperdense areas (650–850 HU) are noted to be located in the sublingual space	Tumor excision	Tumor shows large central area of osseous tissue, characterized by branching and unifying trabeculae of osteoid. At the periphery, more adipose tissue is admixed with areas of bony trabeculae noted.
This case	47	M	Parotid region (L.)	A hard barely mobile mass in left parotid region	Tumor was located at intraparotid gland	10	2.5 × 3.2	MRI scanning shows a well-defined oval lipomatous mass with high signal intensity on T2-weighted image and suppressed signal intensity on fat-suppressed T1- and T2-weighted image. The dense, ossifying layer circumscribing the fatty core appeared as a hypointense cortical line	Tumor excision and partial parotidectomy with facial nerve preservation	Microscopic examination of paraffin embedded section reveals that the tumor exhibited abundant adipose tissue with bony structures

N/A: Not available, M:Male, F:Female, L: Left, R: Right

However, osteolipomas demonstrate various clinical manifestations, including painless soft/hard mobile mass or immovable mass. Even worse, osteolipomas might share overlapping clinical and radiological characteristics with malignant tumors as well as benign neoplasms<sup>17</sup>. Without additional examinations, these clinical presentations are difficult to narrow differential diagnosis and most often mistaken for pleomorphic adenoma, Warthin tumor or malignant tumors. It has been implicating that MRI and CT examination can be used to further characterize osteolipoma, and provide the best tissue definition and allowing visualization of the tumor boundary from surrounding tissue<sup>18,19</sup>. As well known, MRI is the preferred method for visualizing soft tissue neoplasms, whereas CT is helpful for detecting bony structures or ossification. On CT scanning, hypodense fatty tissue with irregular hyperdense ossifying areas might be observed in osteolipoma. MRI imaging of osteolipoma shows well-defined mass with high signal intensity on T1-weight imaging but suppressed signal intensity on fat-suppressed T2-weight images. Therefore, osteolipoma might be diagnosed based on the radiographic findings though combination MRI and CT examination. In our case of osteolipoma, the tumor demonstrates high signal intensity on T2-weighted image and suppressed signal intensity on fat-suppressed T1- and T2-weighted image. A hypointense line was also observed on MRI imaging, indicating that the fatty core mass was circumscribed with an osseous layer. These findings supported the mass can be definitively diagnosed as tumors of adipocytic origin.

So far as now, the pathogenesis of osteolipomas is still not clarified. Some researchers suggested that osteolipomas might be derived from the bidirectional differentiation of multipotent mesenchymal stem cells, which claimed that a lipoma with ossifying tissue is the result of lineage differentiation into both bone and adipose tissue from mesenchymal stem cells<sup>20</sup>. The other widely proposed theory is the metaplasia of fibrous elements into bone tissue from the pre-existing lipoma, resulting from repetitive trauma, metabolic changes or ischemia<sup>1</sup>. Aside from the two hypotheses described above, it has also been implicated that the development of osteolipomas might result from the transformation of fibroblasts into osteoblasts induced by growth factors released from monocytes<sup>21</sup>, or perhaps the ossification due to an inadequate nutritional supply within the center of a large lipoma<sup>14</sup>.

The treatment of osteolipoma is similar with lipoma and tumor excision is recommended. The recurrence is rare after complete excision. As evident in Table 1, the simple tumor excision was used to manage the osteolipoma from salivary gland regions except for the parotid region. All the submandibular and sublingual glands were not damaged and still kept intact in these cases. However, it remains under controversy for the management of parotid gland osteolipoma, as well as lipoma. Several methods have been proposed for managing the parotid lipoma, including enucleation or excision with a small border of parotid gland for encapsulated intra or paraparotid lipomas, superficial parotidectomy and total parotidectomy<sup>10,22,23</sup>. Unsurprisingly, superficial parotidectomy is preferred by most authors. As noted in our case, the tumor was resided at the superficial lobe of parotid and closely attached to facial nerve. MRI examination suggested that the internal boundary of osteolipoma was involved in parotid gland. To preserve the function of parotid gland and remove the tumor completely, we performed partial parotidectomy and excise the tumor but preserve most of parotid gland and facial nerve. In the case reported by Diom

et al., total parotidectomy with facial nerve preservation was employed to treat the osteolipoma due to the mass located between the deep lobe of the gland externally and the parapharyngeal space internally. Taken together, we recommended that tumor excision should be used to manage parotid osteolipoma. To remove tumor completely and preserve the function of facial nerve and healthy parotid gland, the tumor location and its relationship with parotid must be taken into considerations during surgery.

## Conclusion

Osteolipoma is an extremely rare lesion in salivary gland region, which should be considered along with common salivary gland tumors in the differential diagnosis. CT and MRI can accurately characterize osteolipoma and should be utilized to define the size, location, and extent of the neoplasms prior to surgery. Definitive diagnosis can only be achieved with histopathologic examination. Tumor excision is recommended for managing the salivary osteolipoma. However, partial, superficial or total parotidectomy might be required for treating parotid osteolipomas dependent on the tumor location and its relationship with parotid gland.

## Declarations

## Ethics approval and consent to participate

Not applicable.

### Consent for publication

The patient provided informed consent to publish this all presentations of case reports.

### Availability of data and materials

The data and material supporting the conclusions of this article are included within the article.

### Competing interests

The authors declare that they have no conflict of interest

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## Authors' contributions

CW conceived this report and revised it. JH, NX and ZZ drafted the manuscript. NX drafted the pathological part of the manuscript and revised it. All authors read and approved the final manuscript.

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Not applicable

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## Figures

Figure 1

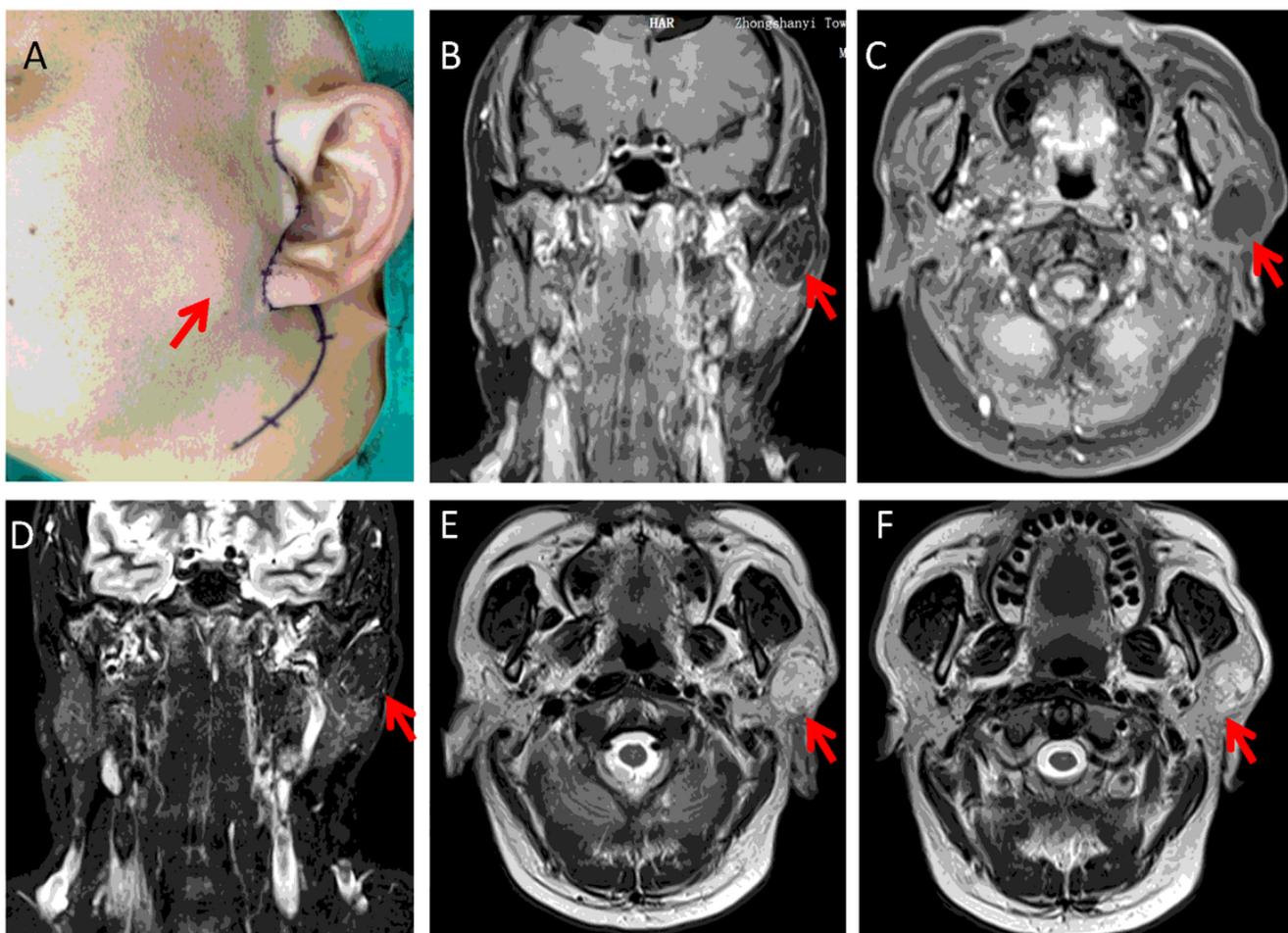


Figure 1

Clinical and MRI imaging revealed an oval mass located at the left parotid gland. A barely mobile mass with a smooth surface was noted in the left cheek (A). A well-defined oval mass was observed in the superficial lobe of left parotid gland with suppressed signal intensity on fat-suppressed T1- (B and C) and fat-suppressed T2-weighted image (D). The mass showed a high signal intensity on T2-weighted image, which was circumscribed with a hypointense line (E). The internal boundary of tumor with parotid gland is indistinct (F).

Figure 2

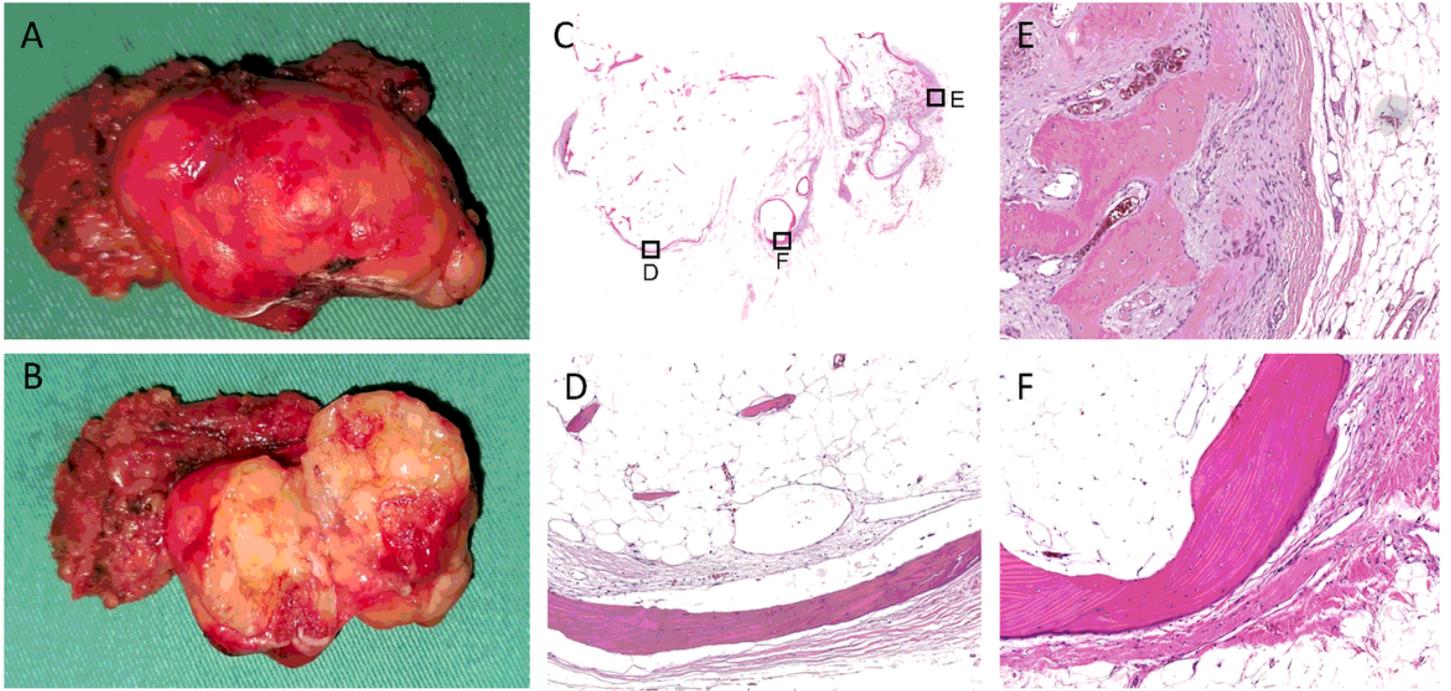


Figure 2

Osteolipoma was diagnosed based on the pathological examination of resected specimen. The resected specimen was a well circumscribed and yellowish mass with hard consistency in general (A). On sectioning, the mass showed a yellow soft tissue with bony structures (B). Microscopic examination revealed the tumor was comprised by mature adipose tissue admixed with areas of bony trabeculae, along with intervening fibrous connective tissue, few inflammatory cells and blood vessel (C: 0.3x, D-F: 10x).