Parosteal lipoma of the mandible: A case report and review of the literature

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Abstract

Introduction: Lipomata are soft-tissue mesenchymal neoplasms that are benign in nature and often asymptomatic. Lipomata commonly occur in the body, however, only 1%–5% are identified within the oral cavity, and lipomata with osseous metaplasia account for <1% of all lipomata. This report describes a patient presenting with an ossifying lipoma with parosteal localization at the mental protuberance. This is followed by review of the current literature for this rare entity.

Materials and Methods: A PubMed literature review designating search terms including "oral" and "parosteal lipoma" or "lipoma with osseous metaplasia" or "ossifying lipoma" or “osseous lipoma” was conducted.

Results: The literature review identified two previous reports of parosteal localization of lipomata in the mandible and 22 cases of lipomata with osseous metaplasia within the oral cavity.

Conclusions: With the initial suspicion of a more malevolent diagnosis, this rare entity, with only two other cases identified, is important to consider in the clinician’s differential diagnoses.

Keywords: Parosteal, lipoma, osseous lipoma, oral

INTRODUCTION

Lipomata are currently considered the most common benign soft-tissue mesenchymal neoplasm. As the main constituent of lipomata is mature adipose tissue, they appear most commonly in areas of the body where adipose tissue is present.[1] 15%–20% of lipomata are found in the head-and-neck region, most often the neck, with only 1%–5% of all lipomata within the oral cavity.[2] Lipomata account for only 0.1%–5% of all benign oral tumors.[3] Lipomata most frequently develop in patients over 40 years of age, more typically in men than women and usually with a slow progression over multiple years.[4,5] Oral lipomata are most frequently detected in the lips, buccal mucosa, lingual and sublingual tissues.[1]

Clinically, lipomata tend to present as well-circumscribed masses, and those involving the oral cavity are frequently yellowish, soft and nontender. Histologically, the main constituent of lipomata is mature adipose tissue without evidence of nuclear or cellular atypia.[1,6]

The case to be discussed is an osteolipoma with parosteal localization, also coined a parosteal lipoma. As far as our knowledge, there are only two previous reports of parosteal localization of lipomata in the mandible, and 22 cases of lipomata with osseous metaplasia within the oral cavity. With the initial suspicion of a more malevolent diagnosis, this rare entity is important to consider in the clinician’s differential diagnoses.
research can identify, this is the third case of a parosteal lipoma of the mandible documented in English-language literature, but it varies in its initial symptom presentation when compared with other reports of lipomata, osteolipoma and parosteal lipomata.

CASE REPORT

A 48-year-old male was referred by his general medical practitioner to the Oral and Maxillofacial Surgery Department at Bristol Dental Hospital, via an expedited suspected cancer pathway, for assessment of a lump of his right lower jaw noted by the patient 3 weeks prior. This lump had visibly enlarged over those few weeks and was becoming painful. The patient was also experiencing constitutional symptoms in this time period, with night sweats, fevers and chills. He had noted a slight facial asymmetry, but had no appetite, weight or skin changes, nor any sensory disturbance.

The patient’s past medical history included hypogonadotropic hypogonadism and gynecomastia, primary lumbar microdiscectomy and gastroesophageal reflux disease. His regular medication included omeprazole, naproxen, testosterone injections and tramadol. The patient had no known allergies and had not smoked cigarettes for 10 years.

On examination, there was a 2 cm palpable bony hard mass fixed to the right mental protuberance, resulting in a subtle facial asymmetry. There was no associated tenderness, no overlying skin changes, sensory deficit or trismus. On intraoral examination, there was no palpable or visible buccal expansion of the labial sulcus, as this lump was positioned close to the inferior mandibular rim. No dental pathology was identified.

RADIOGRAPHIC EXAMINATION

An orthopantomogram and a mandibular occlusal radiograph were taken at this first appointment to assess the composition and location of this bony mass and to identify any potential odontogenic source.

The mandibular true occlusal radiograph [Figure 1] demonstrates a calcified, slightly multilocular appearing lesion arising from the buccal cortex of the mandible in the right parasympyseal region. The occlusal radiograph demonstrates the area as prominent with a mildly irregular periphery and lies adjacent to the lower right lateral incisor to first premolar. Although this lesion was not readily apparent on the panoramic radiograph, it did serve to help steer the differential diagnoses away from odontological causes [Figure 2]. Although features were not entirely consistent with a primary bone malignancy, an urgent biopsy was recommended by the radiologist.

Given the reported rapid progression of the lesion and constitutional symptoms, the working diagnosis was osteosarcoma. Other differential diagnoses at this time included exophytic bony lesions, such as exostosis, osteoma, osteochondroma and chondrosarcoma.[7]

A biopsy was conducted under local anesthetic, at this initial consultation appointment following radiographic investigation. An intraoral incision was made in the right labial vestibule, and the bony lump was evident subperiosteally, but free of the mandibular cortex. Due to well-circumscribed and encapsulated nature of lesion, the base of the lump was dissected easily from the surrounding tissue and excised whole. It did not appear attached to adjacent bone, but extended inferiorly, resting against and below the inferior border of the mandible. There was a small depression of mandibular cortex at the surgical site, suggesting a compression defect from this growth.[8]

HISTOPATHOLOGY

The hard tissue specimen measured 25 mm × 15 mm × 12 mm and was decalcified in formic acid. On microscopic examination, the lesion showed a relatively well-circumscribed polyloid structure, predominantly with mature adipose tissue and lobules of adipocytes. No significant cellular atypia, variation in cell size, nuclear atypia or conspicuous mitotic activity was identified. Islands of mature lamellar bone were scattered throughout the fatty tissue component. There are multiple foci of osseous metaplasia showing trabeculae of remodeling bone, focally incorporating loose fibrofatty tissue with no evidence of hematopoietic cells [Figure 3 and 4].
The diagnosis agreed with the histopathologist was a benign adipocytic tumor of right mandibular parasympysis, with a final differential diagnosis of either parosteal lipoma or lipoma with osseous metaplasia, to be correlated against the clinical picture. As this mass was firm and immobile on clinical examination, with evidence of cortical depression of the underlying mandible, coupled with the knowledge that parosteal lipomata often undergo osseous metaplasia, the diagnosis of parosteal lipoma was made.[8]

Plan for routine 6-month follow-up, but in the COVID-19 pandemic, face-to-face follow-up has been delayed.

DISCUSSION AND REVIEW OF LITERATURE

Lipomata have been separated into four forms by the World Health Organization 2002 guidelines, according to the origin of their location. These forms include: superficial lipoma (arises within subcutaneous tissue); deep lipoma (arises within deep soft tissue, deep to investing fascia), intramuscular or intermuscular lipoma (most commonly within large muscles of thigh, upper arm and shoulder) and parosteal lipoma (arise within the surface of bone, subperiosteally).[5,9] Furthermore, when the lipoma is intimately related to or within bone, they are categorized in accordance with their relation to the parent bones, as intraosseous (within bone), cortical (relating to cortex of bone) or parosteal. Parosteal lipomata are an uncommon type of lipoma and occur in intimate association with the periosteum of bone, mostly affect the long bones and presentation in the mandible is rare.[8] Only two other cases of parosteal lipomata of the oral cavity have been reported, both occurring in the mandible.[8,10]

Osseous changes in lipomata are rare, seeming to occur in some longstanding and large lipomata. Titles, often used interchangeably, for lipoma with osseous components include: osteolipoma; lipoma with osseous metaplasia; osseous lipoma or ossifying lipoma. This subtype accounts for approximately 0.3% of all lipomata. Osteolipomata are found most frequently at the long bones but have been reported at the femur, radius, humerus, tibia, fibula, clavicle, pelvis and oral cavity.[2,8]

Raghunath et al. conducted a review of English literature, identifying a total of 21 cases of osteolipomata of the head and neck, with only twelve cases of osteolipoma presenting in the oral cavity. Osteolipomata were found in the tongue, sublingual tissues, buccal vestibule, buccal mucosa, hard and soft palate and submandibular region.[2]

The parosteal lipomata exhibits a contactual relationship to the periosteum, with commonly an attachment to the periosteum, resulting in an osseous reaction occurring within the tumor. Parosteal lipomata may rest directly on the cortex, with or without cartilage or bone elements contained within the tumor.[2] Parosteal lipomata often lead to definite bony alterations of the parent bones, such as bony hyperostosis, protuberance erosion and compressive changes. Branch-like or linear cortical protuberances and ossification are frequently seen but aggressive bone destruction is consistently absent.[8] Parosteal lipomata usually present morphologically as homogenous, lobulated and often adherent to adjacent bone.[8]

Two main theories for the provenance of an osteolipoma include fibroblasts within the lipoma undergoing metaplasia into osteoblasts, a process which may be initiated by repetitive external microtrauma or ischemia secondary to outgrowth of the tumor's blood supply, leading to modification of the mesenchymal cells. The other theory is that the adipocytes and osseous components originate from a multipotent, undifferentiated mesenchymal stem cell.[1,2,8] Presentation of exophytic osseous components from the cortex of the mandible can highlight suspicions of osteochondroma, osteoma, osteosarcoma or chondrosarcoma. It is important to note that these tumors usually present without surrounding fat components. However, if spiculated periosteal new bone formation with an ill-defined border is present, one should consider osteosarcoma in their differentials.[8,11,12]

A literature review on the search engine PubMed and Google Scholar was conducted in 2020–2021, including search terms with Boolean operators for “oral” and “parosteal lipoma” or “lipoma with osseous metaplasia” or “ossifying lipoma” or “osseous lipoma.”

INCLUSION CRITERIA
- Articles of English-language literature
- Lipoma with osseous change or parosteal lipoma
- Presentation of lipoma in the oral cavity.

EXCLUSION CRITERIA
- Osteolipomata presenting elsewhere on head and neck that didn’t involve the oral cavity
- Osteolipoma of the submandibular space detected by neck examination, without intraoral involvement
- Osteolipoma with additional mesenchymal tissue evident, for example, osteochondrolipoma.
- Insufficient information in report to confirm the lipoma meets the inclusion criteria.

The literature review identified two previous reports of parosteal localization of lipomata in the mandible and
Table 1: Literature review using PubMed and Google Scholar searches and previous literature reviews for osteolipomata and parosteal lipoma of the oral cavity

| Author                  | Year | Age (years) | Male/ Female | Site                          | Clinical presentation | Imaging findings | Time present | Histopathology                                                                 |
|-------------------------|------|-------------|--------------|-------------------------------|----------------------|-----------------|--------------|--------------------------------------------------------------------------------|
| Godby et al.[19]        | 1961 | 54          | Male         | Floor of mouth                | Painless and soft    | Radiopaque mass | 1 year       | Mature adipose tissue, cancellous bone, fibrous connective tissue and striated muscle Diagnosis: Sublingual lipoma with ectopic bone formation |
| Hughes[14]              | 1966 | 69          | Male         | Mandibular buccal vestibule   | Painless and soft    | No pathology   | -            | Fat cells with foci of ossification surrounded by fibrous connective tissue Diagnosis: Intra-oral lipoma with osseous metaplasia |
| Steiner et al.[18]      | 1981 | 50          | Male         | Body of mandible: Right mandibular third molar region | Asymptomatic         | Well-defined radiolucency on OPG | -            | Diagnosis: Parosteal lipoma                                                   |
| Allard et al.[19]       | 1982 | 81          | Female       | Mandibular buccal vestibule   | Facial asymmetry,    | Well-defined radiopaque mass with irregular trabeculae on occlusal radiography | 30-40 years   | Homogenous adipose tissue containing fibrous septa and irregular trabeculae of lamellar bone without hematopoietic tissue Diagnosis: Oral lipoma with osseous metaplasia |
| Piattelli et al.[14]    | 2001 | 49          | Female       | Lateral margin of tongue      | Painless and hard    | -               | 8 years      | Mature adipose tissue containing lamellar bone surrounded by a fibrous pseudocapsule Diagnosis: Osteolipoma |
| Castilho et al.[4]      | 2004 | 65          | Female       | Buccal mucosa                 | Painless and soft    | -               | -            | Mature fat cells supported by fibrous septa. Focal areas of woven bone surrounded by fusiform-shaped mesenchymal cells Diagnosis: osteolipoma |
| Saghafi et al.[15]      | 2008 | 68          | Male         | Mandibular buccal alveolar mucosa | Painless and hard    | No evidence of cortical abnormality or influence on the surrounding structures (radiography) | 4 years       | Adipose tissue with foci of lamellar bone surrounded by mesenchymal cells Diagnosis: Osteolipoma Lobules of adipose tissue separated by fibrovascular connective tissue septa and showing the presence of mature bone |
| Gokul et al.[18]        | 2009 | 6           | Male         | Hard palate (associated with cleft of hard and soft palate) | Painless and soft    | Well-defined hypodense lesion with mixed density, showing a well-defined radiodense body (CT) | Congenital (6 years) | Diagnosis: Osteolipoma Mature adipose tissue with scattered trabeculae of lamellar bone Diagnosis: Osteolipoma |
| De Castro et al.[24]    | 2010 | 47          | Female       | Buccal mucosa                 | Facial asymmetry,    | Patchy areas of radiopacity (occlusal radiography) | 10 years      | Diagnosis: Osteolipoma Mature adipose tissue with scattered trabeculae of lamellar bone Diagnosis: Osteolipoma Lobules of mature adipose tissue separated by fibrous septa Randomly distributed trabeculae of mature lamellar bone and foci of woven bone Diagnosis: Osteolipoma |
| Adebiji et al.[19]      | 2011 | 37          | Female       | Hard palate                   | Painless and hard    | Patchy areas of radiopacity (occlusal radiography) | 10 years      | Diagnosis: Osteolipoma Mature adipose tissue with scattered trabeculae of lamellar bone Diagnosis: Osteolipoma Lobules of mature adipose tissue separated by fibrous septa Randomly distributed trabeculae of mature lamellar bone and foci of woven bone Diagnosis: Osteolipoma |
| Hsu et al.[20]          | 2012 | 71          | Male         | Buccal mucosa                 | Painless and hard    | -               | 4 years      | Diagnosis: Osteolipoma Lorelles of mature adipose tissue separated by fibrous septa Randomly distributed trabeculae of mature lamellar bone and foci of woven bone Diagnosis: Osteolipoma |
| Shabbir and Putnam[21]  | 2013 | 58          | Female       | Mandibular buccal vestibule   | 2×2 cm painless and hard mass, Facial asymmetry | Diffuse area right buccal sulcus with calcifications flecks. No obvious involvement with adjacent bone demonstrated on occlusal radiograph | 1 year        | Diagnosis: Osteolipoma Adipose tissue containing thin-walled vessels with intersecting trabecular type bone with surrounding fibrous tissue |

Contd...
### Table 1: Contd...

| Author         | Year (years) | Age | Male/ Female | Site                        | Clinical presentation                                                                 | Imaging findings                                                                 | Time present | Histopathology                                                                 |
|----------------|--------------|-----|--------------|-----------------------------|---------------------------------------------------------------------------------------|---------------------------------------------------------------------------------|--------------|--------------------------------------------------------------------------------|
| Sun et al.[8]  | 2013         | 48  | Male         | Parosteal lipoma of mandible | Slow growing mass on chin associated with occasional numbness right lower lip. 8×6×5 cm mass beneath chin and bilateral mental areas. Buccogingival sulcus shallower | OPG radiograph showed no major bony changes. CT revealed broad-based, well-demarcated mass with fat attenuation beneath the surface with areas of ossification. Exophytic osseous protuberance of mandible and branch-like periosteal thickening | 20 years     | Mostly mature adipocytes with scattered layers of mature bone foci seen Diagnosis of ossifying parosteal lipoma |
| Bajpai et al.[12] | 2014       | 55  | Male         | Hard palate                 | Painless and hard yellowish mass. Size 1.5×1.5 cm                                      | Patchy area of radiopacity on occlusal radiography                               | 4 years      | Bone trabeculae surrounded by lobules of mature adipocytes separated by fibrous septa Diagnosis: Osteolipoma |
| Amaral et al.[11] | 2015       | 51  | Male         | Mandibular buccal mucosa    | Slight facial asymmetry. Painless and hard mass. Size: 2.0×1.5 cm                     | Well-defined hyperechogenic mass with areas of calcification (ultrasonography)   | 3 years      | Central areas of osseous trabeculae and lobules of mature adipose tissue Diagnosis: Osteolipoma |
| Raghunath and Manjunatha[2] | 2015   | 20  | Female       | Floor of the mouth          | Painless and hard yellowish mass. Size: 6.0×6.0 cm                                      | Well-defined, hypodense lesion with irregular hyperdense areas on CT Spherical radiopacity with an irregular trabecular pattern (soft-tissue radiography) | 3 years      | Central areas of osseous trabeculae and lobules of mature adipose tissue Diagnosis: Osteolipoma |
| Omonte et al.[1]  | 2015         | 29  | Female       | Buccal mucosa               | Painless and hard mass. Size 1.5×1.5 cm                                              | Multiple dense homogenous radio-opacities in the left mandibular posterior region on OPG Contrast-enhanced magnetic resonance image showed that the mass mainly composed of adipose tissue | 8 months     | Diffuse proliferation of mature adipose cells was observed, and mature bone tissue was widely distributed within the tumor. In part of these tissues, a lining of osteoblasts was observed, with no lipoblasts Diagnosis: Osteolipoma |
| Raviraj et al.[22] | 2016       | 38  | Female       | Buccal mucosa               | Painless swelling in her left inner cheek, gradually increasing 2×2×3 cm Presenting symptom of trismus and bone-like hard mass was palpated around lower edge of zygomatic bone. Soft-tissue tumor, 66×45×21 mm | CT shows calcified lesion in the right buccal mucosa                               | 10 years     | Diffuse proliferation of mature adipose tissue. Within one area, there was a fibrous connective tissue surrounding piece of vital bone with large fibro-fatty marrow space and prominent blood vessels Diagnosis: Osteolipoma |
| Fukushima et al.[23] | 2016    | 28  | Male         | Coronid process             | Presenting symptom of trismus and bone-like hard mass was palpated around lower edge of zygomatic bone. Soft-tissue tumor, 66×45×21 mm Mass in right buccal mucosa adjacent to 47 | -                                                                            | 10-12 years   | Diffuse proliferation of mature adipose cells was observed, and mature bone tissue was widely distributed within the tumor. In part of these tissues, a lining of osteoblasts was observed, with no lipoblasts Diagnosis: Osteolipoma |
| Firth et al.[24]  | 2017         | 56  | Female       | Buccal mucosa               |                                                                                      |                                                                                   | -            | Partially encapsulated lobules of adipose tissue. Within one area, there was a fibrous connective tissue surrounding piece of vital bone with large fibro-fatty marrow space and prominent blood vessels Diagnosis: Osteolipoma |
Table 1: Contd...

| Author       | Year (years) | Age (years) | Male/female | Site                                     | Clinical presentation                                                                 | Imaging findings                                                                 | Time present | Histopathology                                                                 |
|--------------|--------------|-------------|-------------|------------------------------------------|---------------------------------------------------------------------------------------|---------------------------------------------------------------------------------|--------------|--------------------------------------------------------------------------------|
| Anbinder et al. [25] | 2017        | 46          | Female      | Vestibule of posterior maxilla           | Slight facial asymmetry. Slowly growing since childhood. Wears complete upper denture 2 cm in size | CT demonstrates well-defined hypodense lesion with hyperdense areas in the vestibular area of the posterior maxilla | ~30 years    | Incisional biopsy disclosed compact lamellar bone that surrounded mature adipose tissue. Lesion was attached to maxilla by wide base. Histopathology showed mature adipose tissue interspersed with mature trabeculae of lamellar bone. Lesion surrounded by compact lamellar bone. Diagnosis: Osteolipoma |
| Arantes et al. [26] | 2017        | 60          | Female      | Parasympyseal mandible/submental         | Painless right submental swelling with discrete facial asymmetry. 1.5 cm well-defined, mobile nodular mass | No alteration on OPG. CT with 3D reconstruction demonstrated a hyperdense mass well-circumscribed on the right parasympysis. Central calcified portion measuring 1.2 cm | 5 years      | Nonencapsulated proliferation of mature adipose tissue. Connective tissue septa served separating adipocytes at lobules. Some areas of normal bony trabeculae were observed. Diagnosis: Osteolipoma |
| Attar and Mohammadi [3] | 2020        | 37          | Female      | Hard palate                              | Slow-growing and painless palatal swelling, 3×4 cm in relation to upper left posterior teeth. Normal appearance of overlying mucosa | Occlusal radiograph showed patchy areas of radiopacity | 10 years     | Mature adipose tissue composed of uniform adipocytes, with trabeculae of lamellar bone scattered throughout. Diagnosed as osteolipoma |
| Sharma and Dhillon [27] | 2020        | 35          | Male        | Hard palate                              | Painless round to oval palatal mass with irregular margins, immobile and nontender 4 cm×2.7 cm×0.8 cm in size | Noncontrast CT scan of facial bones demonstrated well-circumscribed, fat-containing, calcified hard palate lesion | 8 years      | Tissue lined by keratinized stratified squamous epithelium. Scattered bony trabeculae with features of osteoblastic rimming were identified. Diagnosed as hard palate osteolipoma |
| Potter J et al. | 2021        | 48          | Male        | Parasympyseal mandible                   | Painful, apparently rapidly enlarging lesion right parasympysis, with concurrent symptoms of night sweats, fever, and chills. Slight facial asymmetry. Immobile 25 mm×15 mm×12 mm in size | Occlusal radiograph demonstrated calcified, slightly multilocular appearing mass arising from buccal mandible in the right parasympyseal region, prominent with mildly irregular periphery. No abnormality detected on OPG | 3 weeks      | Circumscribed polypoid structure, predominantly mature adipose tissue, with lobules of adipocytes. Islands of mature lamellar bone scattered throughout fatty tissue. Multiple foci of osseous metaplasia and no hematopoietic cells. Diagnosis of parosteal lipoma or lipoma with osseous metaplasia |

OPG: Orthopantomogram, CT: Computed tomography

22 cases of lipoma with osseous metaplasia within the oral cavity. This led to a total of 24 cases, which are detailed below.

Table 1 displays the reported cases of oral osteolipomata chronologically, with inclusion of the date of publication, author, patient age in years (Y) and gender (M– Male, F– Female), presentation and location of the lesion, along with radiographic and histopathological findings. The two cases of parosteal lipomata are highlighted in green.

Table 1 demonstrates that the average, mean age at which osteolipomata are detected is 46-years-old, with the duration of lesion being present for the average mean of
The treatment for lipoma, including parosteal and osseous lipoma, is surgical excision. It is not often difficult to dissect a lipoma contained within soft tissue, or a parosteal lipoma abutting the bone. If the lesion has an intimate osseous relationship, is fixed or has altered the parent bone form, surgical excision requires subperiosteal dissection, with potential for more invasive surgical procedures such as an osteotomy or segmental resection of bone, to free the mass from underlying bone. Local recurrence of these benign neoplasms is rare and malignant changes within parosteal lipoma have not been documented.[2,29]

CONCLUSIONS

Osteolipoma of the oral soft tissues are rare, particularly parosteal lipoma. The increasing cases in the literature should remind the clinician and pathologist to keep this entity in mind. Histopathological diagnosis according to the criteria mentioned in this review will help to categorize these rare lesions and further understand their origin. Despite their benign nature and lack of recurrence, it is essential to differentiate these entities from the malignant lesions, including osteosarcoma.[1]

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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