CASE REPORT

Intraosseous mucoepidermoid carcinoma: a review of the diagnostic imaging features of four jaw cases

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The purpose of this case series is to present the common features of intraosseous mucoepidermoid carcinoma (IMC) of the jaws in plain film and CT imaging. Two oral and maxillofacial radiologists reviewed and characterized the common features of four biopsy-proven cases of IMC in the jaws in plain film and CT imaging obtained from the files of the Department of Oral Radiology, Faculty of Dentistry, University of Toronto, Toronto, Canada. The common features are a well-defined sclerotic periphery, the presence of internal amorphous sclerotic bone and numerous small loculations, lack of septae bordering many of the loculations, and expansion and perforation of the outer cortical plate with extension into surrounding soft tissue. Other characteristics include tooth displacement and root resorption. The four cases of IMC reviewed have common imaging characteristics. All cases share some diagnostic imaging features with other multilocular-appearing entities of the jaws. However, the presence of amorphous sclerotic bone and malignant characteristics can be useful in the differential diagnosis.

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Introduction

Approximately 2–4% of all mucoepidermoid carcinomas arise within the jaws, specifically the molar regions, with the mandible affected three to four times more frequently than the maxillae.¹ Intraosseous mucoepidermoid carcinoma (IMC) is the most common intraosseous salivary gland neoplasm, outnumbering all other types of benign and malignant salivary gland neoplasms combined.²

The published diagnostic imaging features of IMC are often described as a well-defined, unilocular or multilocular, osteolytic radiolucency, with and without association with impacted teeth.^{3–29} Although there have been numerous case reports of IMC, the radiographic descriptions have been non-specific and not well characterized.^{3,4,28} Kochaji et al's²⁸ detailed analysis of published IMC cases in the English-language literature

concludes that 87% of the reports have missing information on the radiographic aspects. Table 1 highlights our review of the English-language literature for “central mucoepidermoid carcinoma” and “intraosseous mucoepidermoid carcinoma” using the PubMed interface of the Medline database. 23 published reports of IMC had adequate radiographic descriptors for the cases. Furthermore, only 3 of these 23 studies on IMC focus specifically on the radiographic features. In 1998, Inagaki et al²³ presented six cases of IMC, but did not detail the radiographic internal structure of the lesions. Johnson and Velez¹⁶ reported one case that contained internal dense radiopacities. Raut and Khedkar²² in 2009 documented a case of IMC that had a multilocular internal structure, thick sclerotic borders and the ability to displace surrounding anatomical structures.

The aim of this review of a case series is to characterize and present the common diagnostic imaging features of IMC of the jaws in plain film and CT imaging.

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Table 1 Number of published intraosseous mucoepidermoid carcinoma cases with adequate radiographic descriptors

<i>Radiographic features</i>	<i>Number of cases</i>	<i>Reference</i>
Well-defined periphery	15	3, 5, 6, 10, 11, 13, 19, 22, 24, 26, 28, 30
Ill-defined periphery	14	12, 14, 23, 29
Unilocular radiolucency	18	3, 5, 6, 10, 11, 21, 23, 24, 25, 26, 28
Multilocular radiolucency	16	13, 17, 19, 21, 22, 23, 25, 27
Internal amorphous sclerotic bone	1	16
Cortical expansion	7	6, 13, 15, 20, 22, 23
Cortical perforation	7	11, 19, 23, 28

Table 2 Demographics of the four cases of intraosseous mucoepidermoid carcinoma

<i>Case number</i>	<i>Age of patient (years)</i>	<i>Sex of patient</i>	<i>Site of lesion</i>
1	51	Female	Right posterior maxilla (from first molar to tuberosity)
2	73	Female	Left posterior mandible (from first molar to mid-ramus)
3	29	Male	Left posterior mandible (from second molar to retromolar trigone)
4	42	Male	Left maxilla, crosses midline (from left tuberosity to right central incisor)

Materials and methods

Four cases of biopsy-proven IMC were retrieved from the files of the Department of Oral Radiology, Faculty of Dentistry, University of Toronto, Toronto, Canada. Of these four cases, three had complete CT studies, and one had a series of plain films that consisted of panoramic, periapical, lateral occlusal and specimen radiographs. The last case also had a haematoxylin and eosin (H&E)-stained section of the resected specimen. Two observers (oral and maxillofacial radiologists) reviewed the imaging studies using the following parameters as a guideline in the analysis: site, epicentre, periphery, shape, internal structure and effects on surrounding structures. Any differences in the findings were resolved by repeating the review to arrive at a consensus.

Results

Patient demographics and sites of the four cases of IMC are presented in Table 2. The study sample consists of two females and two males, with ages between 29 and 73 years at the time of presentation. All four cases originated within the jaw bone and fulfilled the widely accepted diagnostic criteria of IMC: (1) presence of a radiographic distinct osteolytic lesion; (2) positive mucicarmine staining; (3) clinical and histological exclusion of a metastasis or an odontogenic lesion; (4) exclusion of the origin from a soft-tissue salivary gland; (5) histological confirmation.^{3,12,16,21,25,29} Although cortical plate integrity is traditionally a diagnostic criterion of IMC, central origin within bone can be inferred in the absence of a prominent peripheral soft-tissue component despite cortical perforation.^{21,29} All four cases involved the molar region of either the maxilla or mandible, with Case 4 having extensive anterior extension across the maxillary midline.

The common diagnostic features of the four cases of IMC are presented in Table 3. All cases have a well-defined, sclerotic periphery and mixed radiolucent–radiopaque internal structure (Figures 1 and 2). The internal pattern consists of randomly dispersed foci of amorphous sclerotic bone, which appears as irregular masses with cortical bone-like attenuation without any organization into trabeculae or septa, and multiple small radiolucent loculations measuring less than 8 mm with and without coarse bony septal borders. In all four cases, the outer cortex of either the maxilla or mandible is expanded and perforated with extension into surrounding soft tissue. Additional effects on surrounding structures include superior displacement and breaching of the floor of the maxillary sinus in Case 1 (Figure 1b); displacement of the mandibular canal and adjacent molars, and external resorption of the distal root of the second molar in Case 3 (Figure 3a,c); and buccal displacement of the adjacent molar in Case 4 (Figure 4b).

All four cases of IMC are histologically low grade. For Case 3, an H&E-stained section of the specimen was available for comparison with the specimen radiograph. Radiographic–pathological correlation shows that the radiolucent loculations bordered by radiopaque sclerotic septae correspond to cystic components of the tumour bordered by residual bone on the photomicrograph (Figure 3d).

Table 3 Common diagnostic imaging features of the four cases of intraosseous mucoepidermoid carcinoma

<i>Radiographic findings in plain film and CT imaging</i>
Well-defined sclerotic periphery
Amorphous sclerotic bone internally
Multiple small loculations internally
Loculations with and without peripheral septa
Expansion and perforation of the outer cortex with extension into surrounding soft tissues

Figure 1 (a) Axial and (b) coronal. CT bone windows of Case 1 show the epicentre of the lesion at the root apex in the right maxillary alveolar process. The lesion has a well-defined, sclerotic border and a mixed radiolucent–radiopaque internal pattern. The radiopaque pattern consists of multiple, small loculations measuring less than 8 mm, a few of which are bordered by sclerotic septae (straight arrow), and many of which lack such borders (curved arrow). The internal structure also consists of amorphous sclerotic bone (arrowhead). The palatal cortex is expanded and perforated with soft-tissue extension. The floor of the maxillary sinus is superiorly displaced and breached

Discussion

Intraosseous mucoepidermoid carcinomas are rare neoplasms. They are not likely to be the first entity that comes to mind in the differential diagnosis of multilocular lesions of the jaws.^{1,2,30–34} To date, only one case series²³ and two case reports^{16,22} have focused strictly on the diagnostic imaging characteristics of IMC.

Figure 2 Axial CT bone window of Case 2 shows a well-defined sclerotic lesion in the left mandibular body with a sclerotic periphery (straight arrows). The internal pattern is very sclerotic and consists of multiple small loculations bordered by sclerotic septae (arrowhead). These small loculations are in turn surrounded by larger loculations. The buccal cortex demonstrates cystic expansion and perforation (curved arrow)

The present study provides a detailed review of the diagnostic imaging characteristics of IMC in both plain film and CT imaging. The findings of this review confirm the presence of sclerotic bone reported in the published case by Johnson and Velez,¹⁶ and reveal that this was a common characteristic in all four cases. Although Johnson and Velez¹⁶ speculated that bone may be produced by IMC, definitive evidence and consensus on whether IMC has the ability for bone formation is lacking.^{1,2} The masses of sclerotic bone may represent trapped and remodelled bone, or stroma-induced bone formation. The radiolucent loculations with and without bony septae may represent cystic components of the neoplastic islands on a histopathological level. The loculations without bony septae do stand out against the surrounding soft tissue, probably because of the lower X-ray attenuation of fluid than a slightly higher attenuation for the solid, cellular components of the tumour.

All four cases of IMC reviewed here have a well-defined, sclerotic periphery and mixed internal structure, consisting of a multilocular pattern with cystic radiolucencies and coarse bony septae. These cases show a mixture of benign and aggressive behaviour, ranging from tooth displacement and root resorption to more aggressive perforation of outer cortical boundaries and extension into surrounding soft tissue. The multilocularity of IMC and its benign and aggressive growth characteristics are similar to other benign multilocular lesions that can have multiple cyst-like components internally. These would primarily include ameloblastoma, glandular odontogenic cyst and keratocystic odontogenic tumour. However, the lesion with imaging characteristics that most resemble those of IMC would be ameloblastoma.

Ameloblastoma is often described as a well-defined, multilocular, expansile lesion of the jaws.^{30–34} However, the well-defined periphery of ameloblastoma is commonly uniformly thin and corticated, unlike the findings of this review of IMC and the case reported by Raut and Khedkar,²² of an uneven, thick, sclerotic

Figure 3 (a) Panoramic film, (b) lateral occlusal film, (c) specimen film and (d) photomicrograph of a haematoxylin and eosin-stained slide of a section of specimen (2X). Case 3 shows a well-defined, sclerotic border surrounding a mixed radiolucent–radiopacity that consists of amorphous sclerotic bone (straight arrow) and multiple loculations with (arrowhead) and without (curved arrow) sclerotic border. The mandibular canal, the second and third molars are displaced. The distal root of the second molar is resorbed. The section of specimen shows cystic components of mucoepidermoid carcinoma invading the cancellous bone in the mandible. The residual cancellous bone (black straight arrows) between the neoplastic elements likely represents the sclerotic septae (arrowhead) dividing the loculations on film

border. Internally, most loculations in ameloblastoma are characteristically rimmed by multiple bony septae,^{30,34} whereas most loculations of IMC are bordered not by bony septae but by soft tissue of a higher attenuation than the loculations. Also, when present, the septae of IMC are very thick and coarse and less defined, unlike ameloblastoma. Another consistent radiographic feature of IMC in this study, also noted in the case series by Johnson and Velez,¹⁶ is the presence of internal foci of amorphous sclerotic

bone, a finding not typical of solid, multicystic ameloblastoma.^{30–34} However, the desmoplastic variant of ameloblastoma and cases of recurrent conventional ameloblastoma have been documented to contain a similar radiographic internal structure to IMC,^{30,34} and may prove difficult, if not impossible, to distinguish from IMC (Figure 5). The typical behaviour of the IMC seen in this review is to break through the outer cortex of the jaw and extend into surrounding soft tissue. This aggressive characteristic, however, when

Figure 4 (a) Axial and (b) coronal CT bone windows of Case 4 show a well-defined mixed radiolucent–radiopacity with a sclerotic periphery in the left maxilla that has crossed the midline. The internal amorphous sclerotic bone (straight arrow) and loculations are once again demonstrated. A few of the small loculations are not bordered by sclerotic septae (arrowhead). The buccal cortex exhibits considerable cystic expansion and perforation (curved arrow). The lesion has also displaced the molar buccally

Figure 5 Axial CT bone window of a recurrent ameloblastoma of the posterior left maxilla shows similar imaging features of internal amorphous sclerotic bone (straight arrow) and small loculations (curved arrow) without and with sclerotic septae (arrowhead), when compared with the cases of intraosseous mucoepidermoid carcinoma presented

coupled with the sclerotic internal structure, should indicate the need to include IMC in the differential diagnosis.

Two other entities with some similarities to IMC that should be considered in the differential diagnosis are glandular odontogenic cyst and keratocystic odontogenic tumour. Both keratocystic odontogenic tumour and glandular odontogenic cyst may have multiple

cystic radiolucent regions similar to conventional ameloblastoma as the loculations are usually bordered by bony septae. Also, neither entity typically has regions of sclerotic bone. A comprehensive list of multilocular lesions would include odontogenic myxoma, central giant cell lesion, aneurysmal bone cyst (ABC) and haemangioma.^{30,32,34} However, these entities do resemble IMC, as they have other distinguishing radiographic features as follows. Odontogenic myxoma not only has a different internal pattern from IMC, but there is also a common tendency for myxomas to grow along the medullary cavity of the mandibular body with minimal cortical bone expansion.^{30,32,34} Central giant cell lesion and ABC can mimic the expansile nature of IMC but their distinctively low-density, granular bony septae^{30,32,34} differ from the predominantly sclerotic radio-opacities in IMC. Also, ABC, like haemangioma, has a highly characteristic feature of multiple, blood-filled, cyst-like cavities that occasionally produces fluid–fluid levels on CT,^{30,34} and are therefore distinct from IMC. Furthermore, none of these jaw lesions typically has masses of internal sclerotic bone.

In conclusion, this review has highlighted characteristic imaging features of IMC. Since IMC are rare entities, the presence of two common findings, namely internal sclerotic bony masses and perforation of the external cortex with extension into surrounding soft tissue, should alert the oral radiologist to include IMC in the radiographic differential diagnosis of multilocular lesions of the jaws.

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