Radiology Case Reports 12 (2017) 756–759

Available online at www.sciencedirect.com

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

Head and Neck

Peripheral osteoma, compound odontoma, focal cemento-osseous dysplasia, and cemento-ossifying fibroma in the same hemimandible: CBCT findings of an unusual case

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ARTICLE INFO

Article history:
Received 9 June 2017
Received in revised form 4 August 2017
Accepted 20 August 2017
Available online

Keywords:
Osteoma
Odontoma
Cementoma
Fibroma, ossifying
Cone-beam computed tomography

ABSTRACT

Peripheral osteoma is the most common subtype of osteoma that arises most frequently in the craniofacial bones. It may occur at any age with a male-to-female ratio of 2:1. Peripheral osteoma may affect the mandible, particularly the ramus and the condyle. Compound odontoma is a subtype of odontoma that occurs in young subjects without gender predilection. It affects the maxilla more frequently than the mandible. Focal cemento-osseous dysplasia and cemento-ossifying fibroma are 2 benign fibro-osseous lesions with a female predominance that occur most commonly in the posterior region of the mandible. We report the first case involving the simultaneous occurrence of these 4 benign lesions in the same hemimandible diagnosed by CBCT.

Introduction

Osteoma is a benign osteogenic tumor consisting of a mature compact or cancellous bone that may occur at any age (typically between the second and fifth decades); this tumor is more frequent in men than in women, with a male-to-female ratio of 2:1 [1].

Solitary osteomas may be classified as peripheral, central, or extraskeletal [1]. Peripheral osteoma (PO), which is the most common subtype of osteoma, arises from the cortical plate (specifically, the periosteum) [1]. POs are most frequently found in the craniofacial area but are rare in the jaws. The mandible is more often affected than the maxilla; when POs originate from the mandible, the most common location is the posterior region, particularly the angle or the condyle [1]. Radiologically, PO appears as a well-defined, exophytic (pedunculated or sessile) bone density mass attached to the cortical plate [1].

Odontoma is considered by many authors to be the most common odontogenic tumor of the jaws; however, in the

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https://doi.org/10.1016/j.radcr.2017.08.011

literature, the reported prevalence of this tumor ranges from 6.0% [2] to 75.6% [3]. Odontoma has no gender predilection and occurs in younger patients, with peak incidence during the second decade of life [4]. Odontomas are currently considered to be hamartomas, and they are classified as compound or complex, depending on their composition. Compound odontoma (CO) is most frequently found in the anterior region of the maxilla, whereas the posterior region of the mandible is the most common location for complex odontoma [4]. On radiological imaging, CO exhibits multiple small, high-density structures similar to the teeth (denticles), whereas complex odontoma appears as an amorphous hyperdense mass that does not resemble the teeth; COs and complex odontomas are typically surrounded by a low-density rim [4,5].

Focal cemento-osseous dysplasia (FocCOD), which is the most common benign fibro-osseous lesion of the jaw, is a subtype of cemento-osseous dysplasia [5,6]. The peak incidence of FocCOD is between the third and the fifth decades of life, and FocCOD is most common in women, particularly in white women [5,6]. FocCOD may develop in any tooth-bearing area of the jaws but occurs most frequently in the posterior mandible at the periapex of a vital tooth as well as within previous extraction sites [5,6]. The radiological appearance of FocCODs may vary, depending on the stage of lesion maturation [5,6]. Initially, FocCOD may appear as a well-defined osteolytic lesion around the apex of a vital tooth. During this early stage, FocCOD may be confused with a periapical inflammatory lesion (eg, a radicular abscess, granuloma, or cyst); however, such lesions affect nonvital teeth. During later stages, the inner portion of a FocCOD progressively develops a denser component that is typically surrounded by a low-attenuation halo [5,6].

Cemento-ossifying fibroma (COF) is a relatively rare benign, nonodontogenic, fibro-osseous lesion of the mandible. This tumor may occur at any age but is most common between the third and the fourth decades of life, with a predilection for women [5,7]. The majority of COFs are detected in the posterior region of the mandible, most commonly in the molar and premolar areas [5]. Radiologically, COF appears as a solitary, well-defined, unilocular, expansile mass with a corticated border and varying levels of internal density depending on the lesion maturity [5].

In this article, we describe an unusual case involving a nonsyndromic 50-year-old white female patient who was diagnosed with PO, CO, FocCOD, and COF in the same hemimandible following a cone beam computed tomography (CBCT) scan. We also describe the CBCT features of these 4 lesions.

Case report

A 50-year-old woman was referred to the Department of Radiology by the Dental Clinic of our university for the evaluation of a painless slow-growing, intraoral, exophytic submucosal lesion arising from the superior lingual aspect of the right mandibular body. The patient was in good health, her medical history was unremarkable (no previous facial trauma), and there was no evidence of systemic disease or hereditary syndromes.

Fig. 1 – Cone beam computed tomography images of the peripheral osteoma. Axial (A) and cross-sectional (B) views show a well-defined bone density lesion (asterisk) attached by a pedicle (curved arrow) to the cortical lingual plate of the right mandibular body near the second premolar (45). The overlying mucosal and submucosal layers (arrowheads) are lifted by the bone density lesion (asterisk).

On intraoral examination, the lesion was hard, firm, and fixed relative to the mandible. The lesion measured approximately 1 cm in diameter, and the overlying mucosa was normal. No other lesions were observed, and the patient's dentition status was within the normal range. Neck lymphadenopathy was not detected.

The clinical appearance of the lesion was not typical for mandibular torus. Based on the site of origin and the exophytic growth pattern of the lesion, no intraoral periapical or panoramic radiographs were considered to be useful. Therefore, given the characteristics of the lesion and the unavailability of an image receptor for occlusal radiography, a CBCT exam was performed.

CBCT images showed a 1×1 cm pedunculated, exophytic hyperdense lesion arising from the lingual cortical plate of the right mandibular body, near the premolar region (Fig. 1). For this lesion, the clinical features (no previous local trauma) and radiological features on CBCT images were consistent with PO.

The CBCT scan identified another lesion in the same hemimandible, between the root of 45 (the second premolar) and the mesial root of 46 (the first molar), measuring 1.2×0.8 cm and consisting of small toothlike structures surrounded by a thin, low-attenuation rim (Fig. 2). These radiological features were diagnostic for CO.

CBCT images of a slightly more distal region, around the apex of the distal root of 46, showed a 0.5×0.6 cm lesion consisting of a small central hyperdense component surrounded by a low-attenuation halo (Fig. 3). A focal discontinuity in the lamina dura of the distal root of 46 was found near the apex. No signs of root resorption or fusion with the root were observed. The patient's age and the lesion's imaging features were considered to be indicative of FocCOD.

Finally, CBCT images showed a fourth mandibular lesion in the posterior region of the mandibular body, deep at the roots

Fig. 2 – Cone beam computed tomography images of the compound odontoma. Axial (A) and cross-sectional (B) views show a group of small toothlike structures (arrows) surrounded by a thin low-attenuation rim (curved arrows). The lesion is located between the root of the second premolar and the mesial root of the first molar (arrowheads).

of 48 (the third molar). This lesion measured 1.8 × 1.2 cm and appeared as a well-defined expansile lesion with mixed density surrounded by a thick, corticated border (Fig. 4). The lesion induced bone remodeling of the surrounding lingual cortical plate and minimal buccal displacement of the mandibular canal (Fig. 4). No signs of root resorption or tooth displacement were observed. The patient's gender and the lesion's imaging features were considered to be consistent with COF.

No bone tumor lesions were observed in the left hemimandible.

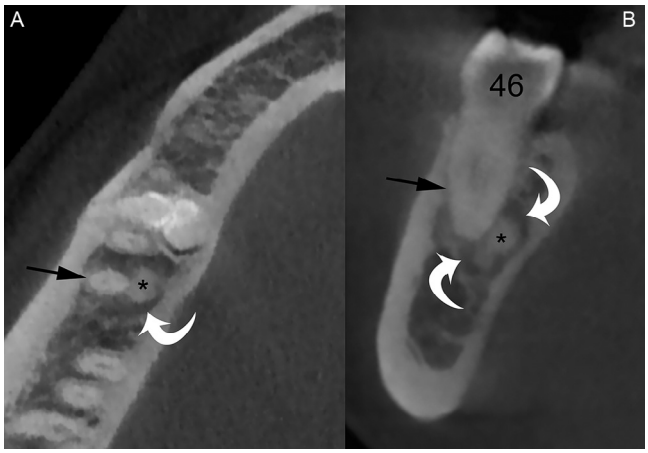

Fig. 3 – Cone beam computed tomography images of the focal cemento-osseous dysplasia. Axial (A) and cross-sectional (B) views reveal a small, well-defined hyperdense lesion (asterisks) surrounded by a low-attenuation rim (curved arrows) located around the lingual aspect of the apex of the distal root (arrows) of the first molar (46). A focal disruption in the lamina dura is observed. No signs of root resorption or fusion with the root are evident.

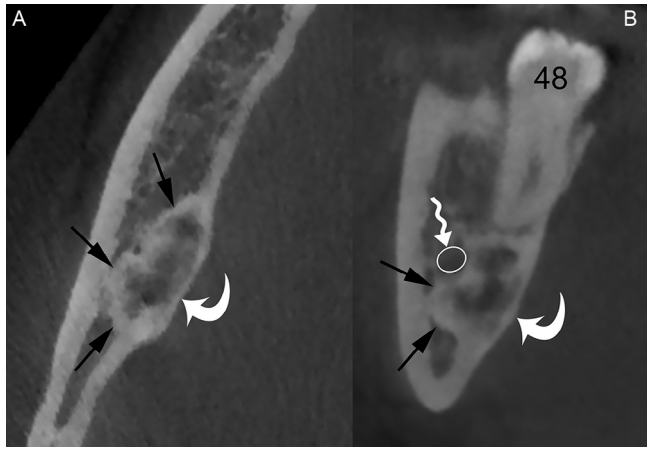

Fig. 4 – Cone beam computed tomography images of the cemento-ossifying fibroma. Axial (A) and cross-sectional (B) views show a well-defined lesion with mixed density surrounded by a corticated rim (arrows) in the posterior region of the mandibular body, deep at the root of the third molar (48). Bone remodeling of the lingual cortical plate (curved arrows) and minimal buccal displacement of the mandibular canal (wavy arrow and circle) are also observed.

Based on CBCT findings, the patient was sent to an oral surgeon. However, considering the natural history of CO (stop growing when normal teeth completed their formation), FocCOD (very limited growth potential), and COF (mandibular lesions exhibit a less aggressive behavior than paranasal sinuses lesions) and the risks associated with excisional biopsy (perioperative hemorrhage and postoperative infection), only the PO was surgically removed due to moderate difficulty in mastication. To date, the patient has undergone a periodic clinicoradiological follow-up and no other surgical treatment has been performed.

Discussion

The medical literature includes several reported cases of multiple benign radiopaque or hyperdense lesions on the jaws [8], especially in patients with Gardner syndrome [9]. However, the occurrence of multiple benign radiopaque or hyperdense lesions with different histologic types in the same jaw is extremely rare.

To our knowledge, this article describes the first case in which the simultaneous occurrence of PO, CO, FocCOD, and COF in the same hemimandible has been observed (Fig. 5). In a literature search of the PubMed database, we found only 1 case report published in 2011 in which the occurrence of 3 different lesions in the same jaw was described [10]. In that report, Hosseini and Moslemi [10] described the case of a 46-year-old Iranian woman who was diagnosed with COF, periapical COD, and complex odontoma in the same mandible. In their case, the clinical and radiographic features of the 3 diagnosed lesions (based on intraoral and panoramic radiographs)

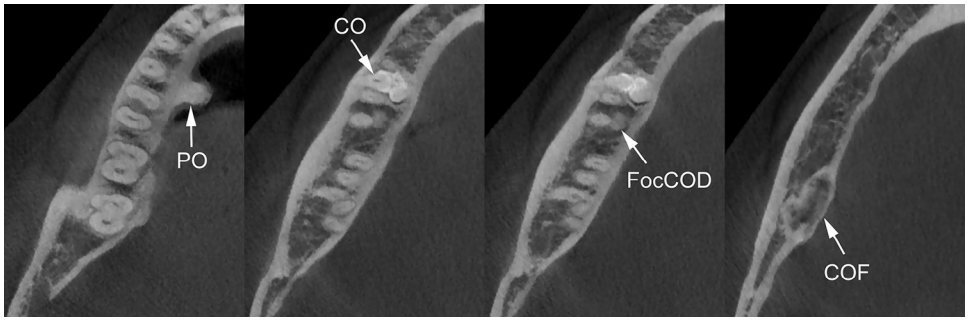

Fig. 5 – Axial cone beam computed tomography images show all 4 lesions of the right mandibular body. PO, peripheral osteoma; CO, compound odontoma; FocCOD, focal cemento-osseous dysplasia; COF, cemento-ossifying fibroma.

were relatively similar to those that are typically observed for such lesions.

Similarly, in our case, radiological findings of the 4 lesions (based on CBCT images) corresponded to typical forms; however, clinical features (patient age, patient gender, and lesion location) differed from those commonly described in the literature for PO and CO.

According to many studies, PO has a predilection for men, and the posterior region, especially the angle and the condyle, is considered to be the most common location [1]. Conversely, in the present case, the patient was a woman and the PO arose from the right mandibular body, near the premolar region.

The literature generally indicates that CO occurs most frequently in younger patients and is most commonly found in the anterior region of the maxilla [4]. In contrast, our patient was a 50-year-old woman, and her CO was located in the mandibular body between the roots of the second premolar and the first molar.

Another aspect that differentiates the present case from that described by Hosseini and Moslemi [10] is that, in our patient, the 4 described lesions were located in the right hemimandible. In contrast, in Hosseini and Moslemi's patient, 1 of the 3 lesions was located on the left hemimandible, and the remaining 2 lesions were on the right hemimandible. Conversely, similarities between our case and Hosseini and Moslemi's case include patient age (approximately 50 years), patient gender (female), the absence of systemic disease or hereditary syndromes, and the 3 types of lesions (COF, odontoma, and COD).

In the case described by Hosseini and Moslemi [10], diagnoses of periapical COD, COF, and complex odontoma were suggested based on intraoral and panoramic radiographs (no CBCT scan was performed) and confirmed with excisional biopsy. In contrast to Hosseini and Moslemi's case, in the currently reported case, diagnoses were based on CBCT features, and no histologic confirmation was obtained. However, it is noteworthy that CBCT images clearly demonstrated the location, the extension, and the internal features of jaw lesions. The accurate morphologic analysis obtained using CBCT images provides significant incremental benefits to the differential diagnosis of jaw lesions. Therefore, we believe that, in the future, the routine use of CBCT could reduce the number of unnecessary excisional biopsies for asymptomatic benign lesions where a decision to defer treatment is the best choice.

In conclusion, the described case is the first reported case in the literature involving the simultaneous occurrence of 4 different radiopaque or hyperdense lesions in the same hemimandible in a nonsyndromic patient. This extremely rare combination may be attributable to chance, as Hosseini and Moslemi suggested [10]; however, it is possible that this incidental occurrence reflects a hormonal imbalance or a genetic disorder in the regulation of bone remodeling [11].

REFERENCES

[1] de França TR, Gueiros LA, de Castro JF, Catunda I, Leão JC, da Cruz Perez DE. Solitary peripheral osteomas of the jaws. Imaging Sci Dent 2012;42:99–103.
[2] Sriram G, Shetty RP. Odontogenic tumors: a study of 250 cases in an Indian teaching hospital. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 2008;105:e14–21.
[3] Buchner A, Merrell PW, Carpenter WM. Relative frequency of central odontogenic tumors: a study of 1,088 cases from Northern California and comparison to studies from other parts of the world. J Oral Maxillofac Surg 2006;64:1343–52.
[4] Iatrou I, Vardas E, Theologie-Lygidakis N, Leventis M. A retrospective analysis of the characteristics, treatment and follow-up of 26 odontomas in Greek children. J Oral Sci 2010;52:439–47.
[5] Curé JK, Vattoth S, Shah R. Radiopaque jaw lesions: an approach to the differential diagnosis. Radiographics 2012;32:1909–25.
[6] Komabayashi T, Zhu Q. Cemento-osseous dysplasia in an elderly Asian male: a case report. J Oral Sci 2011;53:117–20.
[7] Liu Y, Wang H, You M, Yang Z, Miao J, Shimizutani K, et al. Ossifying fibromas of the jaw bone: 20 cases. Dentomaxillofac Radiol 2010;39:57–63.
[8] Sun L, Sun Z, Ma X. Multiple complex odontoma of the maxilla and the mandible. Oral Surg Oral Med Oral Pathol Oral Radiol 2015;120:e11–6.
[9] Fonseca LC, Kodama NK, Nunes FC, Maciel PH, Fonseca FA, Roitberg M, et al. Radiographic assessment of Gardner's syndrome. Dentomaxillofac Radiol 2007;36:121–4.
[10] Hosseini FA, Moslemi E. Central ossifying fibroma, periapical cemento-osseous dysplasia and complex odontoma occurring in the same jaw. Clin Pract 2011;1:e36.
[11] de Noronha Santos Netto J, Machado Cerri J, Miranda AM, Pires FR. Benign fibro-osseous lesions: clinicopathologic features from 143 cases diagnosed in an oral diagnosis setting. Oral Surg Oral Med Oral Pathol Oral Radiol 2013;115:e56–65.