Journal of Dentomaxillofacial Radiology, Pathology and Surgery

Spring 2023, Volume12, Number 2

Case Report: Combination of the Skeletal and Extra-skeletal Craniofacial Osteomas: A Case Report

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Citation: Dalili Kajan Z, Yaghoobi S, Khosravifard N, Hoseini H, Kashi F, Karimian R. Combination of the Skeletal and Extra-skeletal Craniofacial Osteomas: A Case Report. Journal of Dentomaxillofacial Radiology, Pathology and Surgery. 2023; 12(2):36-41 http://dx.doi.org/10.32598/3dj.7.4.145

http://3dj.gums.ac.ir

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Article info:
Received: 2023/ 03/03
Accepted: 2023/08/21

Keywords:
Osteoma
Mandible
Infra-temporal fossa

ABSTRACT

Osteoma is a slow-growing benign osteogenic tumor. Osteomas can be central, peripheral, and extra-skeletal. The prevalence of extra-skeletal osteoma often found in muscles is very low. Osteomas unrelated to the syndrome often occurs as solitary tumors, but sometimes multiple osteomas are unrelated to the syndrome either. Here, we report a rare non-syndromic craniofacial osteoma, a combination of skeletal and extra-skeletal types. The patient complained of dull pain in the right maxillary molar area during mastication. Panoramic and CBCT showed multiple homogenous opacities on the right mandibular body and condylar neck as well as in the right infratemporal fossa and masticator space, having a pressure effect on the base of the skull. A biopsy from the mass on the mandibular body confirmed osteoma. The absence of the other manifestations of Gardner syndrome made the patient a rare case of multiple non-syndromic osteomas.

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Introduction

Osteoma is a slow-growing benign osteogenic tumor created by the proliferation of dense or spongy bone. The prevalence of this tumor is 0.01 to 0.43.[1] It is more common in the skull, maxillary sinuses, other paranasal sinuses, and facial bones. More than 78% of patients are over 40 years old (2).

Osteoma can be central, peripheral, and extra-skeletal. The central osteoma originates from the endosteum, and the peripheral type originates from the periosteum. The prevalence of extra-skeletal osteoma often found in muscles is very low. Most cases have been reported on the tongue and skin. A limited number found in the extremities include the hip, thigh, and hand (3).

Osteomas unrelated to the syndrome often are solitary tumors. Only a limited number of previous studies have presented multiple osteomas unrelated to the syndrome (1,4).

Multiple osteomas could be accompanied by other findings such as intestinal polyps, fibromatosis, impacted or undeveloped teeth, and cysts or skin tumors such as epidermoid cysts. These manifestations may indicate Gardner syndrome, a genetic disorder caused by an autosomal dominant mutation on chromosome 5 (1,2).

Intestinal polyps are adenomatous and are prone to malignant changes in almost 100% of cases. So a dentist plays an essential role in the early diagnosis and referring the patient to an internist for more investigation (5-7). This study reports a rare case of non-syndromic craniofacial osteomas combination of peripheral, central, and extra-skeletal types.

Case report

The present study was approved by the Ethics Committee of Guilan University of Medical Sciences (IR.GUMS.REC.1401.473). Additionally written informed consent was obtained from the patient. A 43-year-old female presented to the maxillofacial radiology department at Guilan University of Medical Sciences with the chief complaint of dull pain during mastication in the right maxillary molar area. She also complains of referral pain to the temporomandibular joint (TMJ), periauricular and temporal regions. She declared that the pain had occurred periodically with the constant pattern and then disappeared after some days. She had no other signs of TMJ problems. The skin over these areas was normal without redness or elevated temperature. She had no significant past medical and dental history except for asthma.

In clinical examinations, the maximum mouth opening was 55 mm. There was no tenderness on the bilateral masticatory muscle during palpation. There was no jaw deviation or clicking sound while opening and closing the mouth. Intraorally, there was no significant abnormal finding except for several dental caries.

Moreover, a bony-hard sessile mass with a 1.5 cm diameter was palpated at the angle of the right mandible. She had been aware of it for more than fourteen years with relatively slow growth.

Panoramic radiography was taken as the first step for radiographic examination. In panoramic view, multiple well-defined radiopacities were observed in the right mandibular body, the posterior portion of the condylar neck, the sigmoid notch, and maybe in the right infratemporal area. (Figure 1) In differential diagnosis, multiple osteomas were considered the diagnosis. In the second step, a CBCT image was taken from the patient.

The CBCT images (axial and coronal views) (figure 2 A-B) reveal:

-A compact homogenous opacities on the buccal aspect of the right mandibular body (1.5x1.5 cm);

- The second one was a cauliflower-like opacity in the right infratemporal fossa and masticator space, having a pressure effect on the base of the skull

- The third one was two adhered opacities in the buccal and medial aspects of the condylar neck of the mandible as a peripheral form

- Expansion and increased internal opacity of upper third of right mandibular ramus that could

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be a cancellous form of osteoma

- The right condylar head is more opaque and more prominent than the counterpart

- Rt glenoid fossa is wider than the normal one, and the dehiscence of the cortical boundary is obvious.

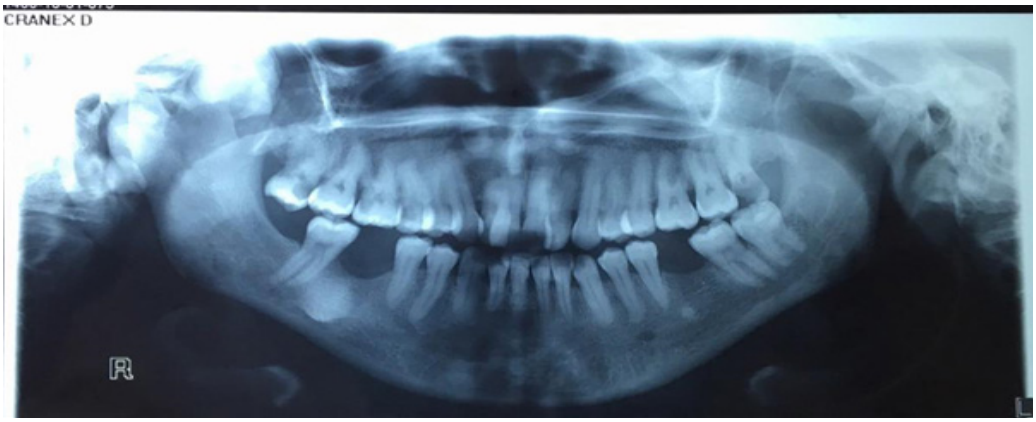
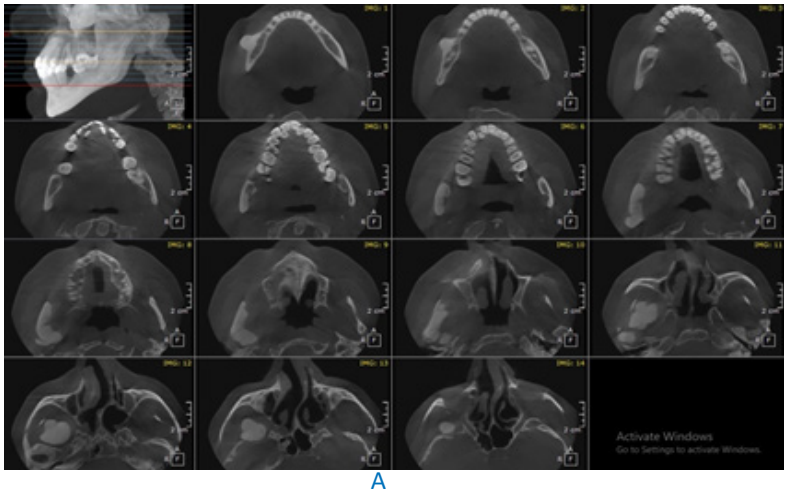
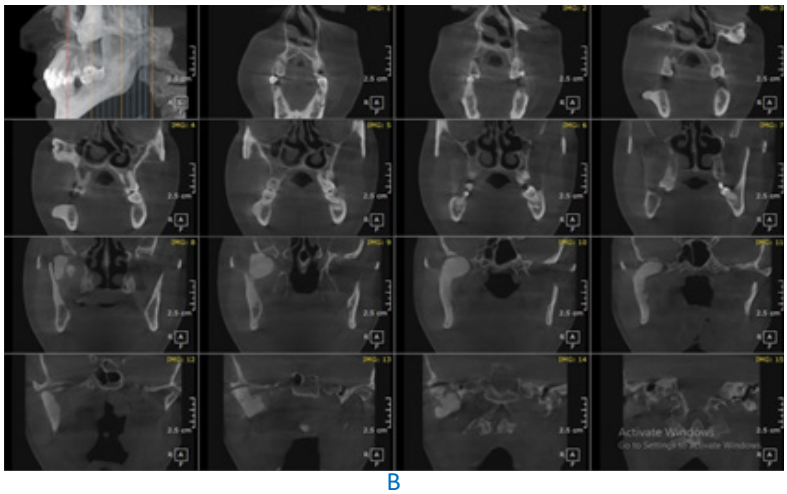


Fig 1. The panoramic view shows multiple well-defined radiopacities in the right mandibular body and the right temporomandibular joint area.



A



B

Fig 2. The CBCT images; A) Coronal and B) Axial views, reveal a homogenous opacity on the buccal aspect of the right mandibular body, a cauliflower-like opacity in the right infratemporal fossa and masticator space with the erosion of the base of the skull, and two adhered opacities in the buccal and medial aspects of the right condylar neck

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The three-dimensional images are also shown in Figure 3(A-D).

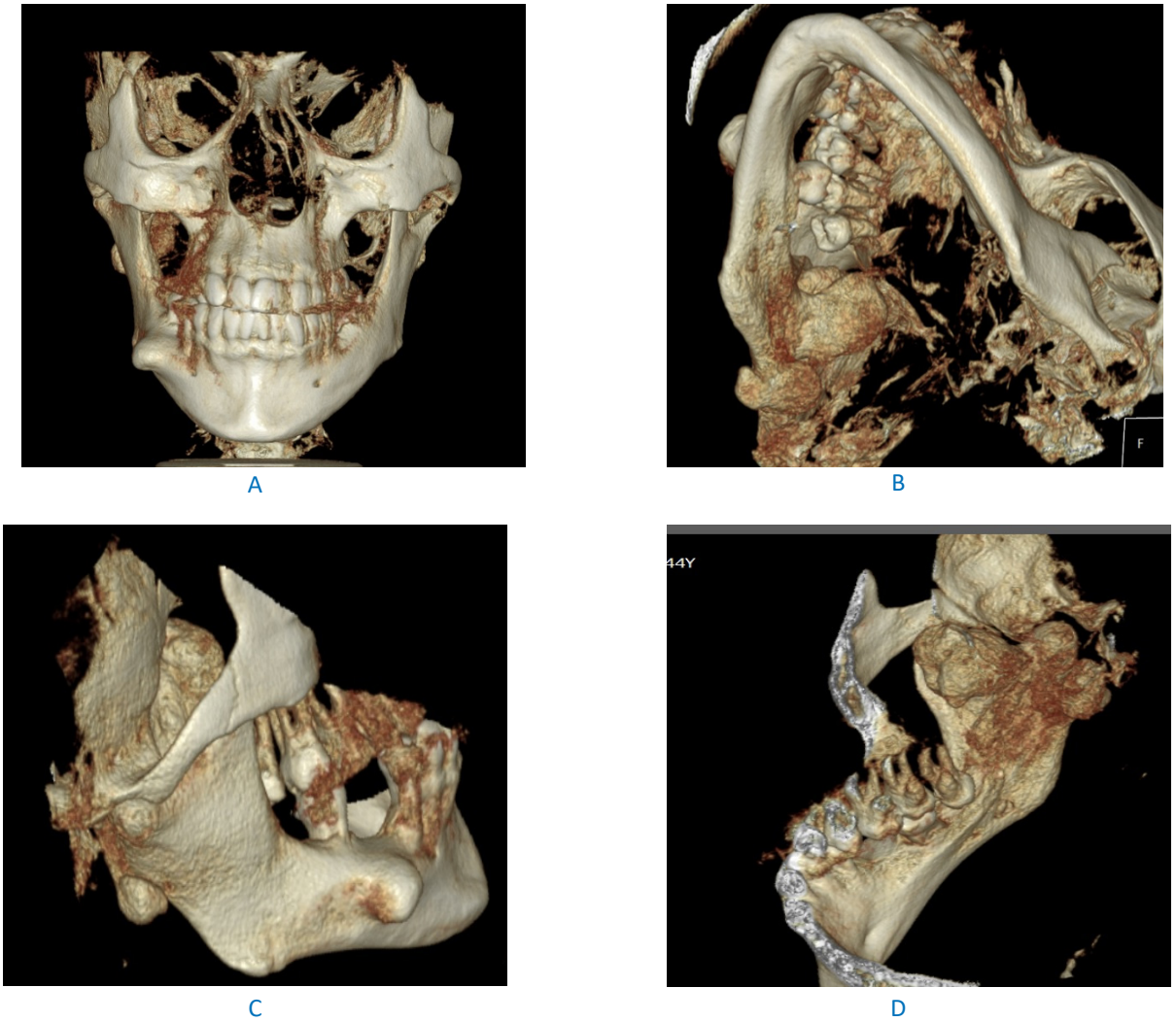

Fig 3. Three-dimensional views of multiple osteomas (A, B, C, D)

A biopsy from the mass on the mandibular body was performed. A microscopic examination revealed dense lamellar compact bone, which confirmed peripheral osteoma (Figure4). These clinical, radiographic, and histologic findings suggested multiple osteomas and the risk of Gardner syndrome. Therefore, the patient was referred to the department of the gastrointestinal department for colonoscopy and, if necessary, biopsy. The colonoscopy revealed nothing except a tiny polyp in the cecum and hyperemia in the terminal portion of the ileum.

Pathological biopsy investigations from the terminal ileum showed chronic ileitis with without granulomas. Additionally, a polyp was detected in the cecum, suggestive of a hyperplastic polyp. The sigmoid mucosa also revealed moderate chronic inflammation with mild subepithelial hemorrhage. So, these findings did not confirm Gardner syndrome.

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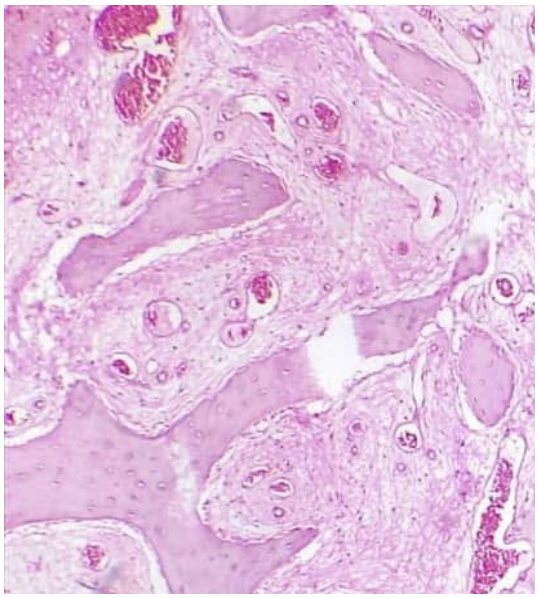

Fig 4. Histopathologic findings shows admixture of mature lamellar and woven bone trabeculae, rimmed by osteoblasts without cytological atypia. Hypocellular well vascularized, loose fibrours stroma is also evident in intertrabecular space.

Discussion

Osteoma is a benign, slow-growing tumor often occurring in the craniofacial region (3). Li et al. found that, excluding osteomas in the maxillofacial sinuses, about 98 solitary osteomas in the mandible and 9 in the maxilla have been reported in English-language literature over the past 80 years, mostly peripheral osteomas (3).

Osteoma is a solitary lesion, and the multiple types are often a part of the clinical manifestations of Gardner syndrome; however, there have been reports of multiple cases of non-syndromic craniofacial osteomas (1,4, and 8) For example, in 2013, Shanavas et al. reported multiple isolated osteomas in the forehead of a 39-year-old patient (8). Yoo et al. reported a similar case in a 72-year-old woman with non-syndromic multiple osteomas in the scalp and forehead in 2020 (1). Olivares et al. presented a rare case of multiple mandibular osteomas in a 36-year-old woman with no syndrome (9). In all of the above reports, osteomas were peripheral. However, based on our knowledge, multiple osteomas in a patient with a combination of peripheral and extra-osseous types are extremely rare and have not been reported until now.

Patients with multiple osteomas should be evaluated for the possibility of Gardner syndrome. These patients may report symptoms such as abdominal pain, gastrointestinal bleeding, or diarrhea. In addition, colorectal polyposis, multiple osteomas, impacted or unerupted teeth, and skin tumors can also be the other manifestations of this syndrome (1). Therefore, the internist performed endoscopic examinations to evaluate the gastrointestinal tract in the present patient. He found a small polyp in the cecum with no malignant or dysplastic transformation. So, the patient was a rare case of multiple non-syndromic osteomas.

In the present case, osteomas were detected in four areas, two of which were in the medial and lateral surfaces of the condylar neck. According to a review study in 2018, from 1927, when condylar osteoma was first described, until 2018, twenty-three cases of condylar osteoma were reported in English-language sources (10).

On the other hand, in the present study, soft tissue osteoma in the infratemporal fossa and masticatory space was associated with skeletal osteomas. Radiographic examinations in the present study showed the pressure effect of the mentioned soft tissue osteoma on the base of the skull, which has not been reported in any of the reports of soft tissue osteoma in the craniofacial region. The reported extra-osseous osteomas usually arise from the oral cavity, skin, or extremities (11,12,13), and in a few cases, they can be found in the head and neck area (3,14]). Li et al. reported osteomas in the pterygomandibular region. They stated that this was the first reported case of soft tissue osteoma in the maxillofacial region that had developed in an area except for the tongue or skin (3). In 2021, Roh et al. reported a rare case of soft tissue osteoma in the temporal area in a 65-year-old woman.(14) Our efforts to find more reports of soft tissue osteoma in the head and neck area were inconclusive.

Conclusion

Therefore, the present case report can be considered a rare case of extra-skeletal osteoma that has been observed in the infra-temporal fossa and masticatory space with the pressure effect on the base of the skull. Also, the combination of extra-skeletal osteoma with skeletal (peripheral and central) type in a patient without syndrome has not been reported in any article.

Acknowledgments

None

Authors' contributions

Zahra Dalili Kajan: Conceptualization, Methodology, Writing - Review & Editing **Soophia Yaghoobi:** Resources, Investigation, Visualization **Negar Khosravifard:** Methodology, Visualization **Hadi Hoseini:** Writing - Original Draft, Data Curation **Faezeh Kashi:** Funding acquisition, Project administration, Supervision **Roghayeh Karimian:** Writing - Review & Editing Resources

Conflict of Interests

The authors declare no conflict of interest.

Ethical declarations

IR.GUMS.REC.1401.473

Financial support

None

Availability of data and material

The datasets used and/or analyzed during the current study are available from the corresponding author on reasonable request.

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