Received: 31 July 2023 | Revised: 18 November 2023 | Accepted: 4 January 2024

DOI: 10.1002/osi2.1228

CASE REPORT

Oral Science International
Official Journal of the Japanese Stomatological Society

WILEY

Mandibular odontogenic cyst atypically extended into the submandibular region with thickened bone formation: A case report

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Funding information
None

Abstract

Background: Odontogenic cysts can cause thinning and expansion of the surrounding cortical bone; however, bone formation extending into the submandibular region, resembling periosteal reactions, is rarely observed in odontogenic cysts.

Case Presentation: A 52-year-old man presented with painful submandibular swelling and dyspnea. Computed tomography revealed an extensive mandibular cystic lesion extending to the submandibular region accompanied by thickened bone formation, mimicking lobulated shell-type periosteal reaction. Percutaneous cystectomy with extensive mandibular bone resection was performed.

Conclusion: Prolonged existence of extensive cystic lesion and inflammatory stimulation may result in bone elongation into the submandibular region, mimicking periosteal reaction.

KEYWORDS
dyspnea, odontogenic cysts, periosteal reaction, submandibular region

1 | INTRODUCTION

The periosteum reacts to several underlying insults.[1] Periosteal reactions can be observed in the jaws in several disorders, including osteomyelitis, trauma, malignancy, odontogenic tumors, and odontogenic cysts.[2] Periosteal reaction is nonspecific and shows various patterns that reflect the intensity, aggressiveness, and duration of the temporal factors of the lesion.[1,3] Although odontogenic cyst enlargement tends to cause cortical bone thinning and expansion, bone thickening owing to periosteal reaction is rarely observed around these cysts. Herein, we report a case of mandibular odontogenic cyst that extended into the submandibular region and was accompanied by new bone formation, mimicking a shell-type periosteal reaction.

2 | CASE REPORT

A 52-year-old man visited the previous medical institution with a chief complaint of painful swelling in the right submandibular region and respiratory distress. He underwent an incisional drainage via submandibular approach at that facility, following which he was referred to our hospital for further examination and treatment. Physical examination revealed a hen's egg-sized mass-like, hard tissue in the submandibular region and swelling in the floor of the mouth (Figure 1). Panoramic radiography revealed a large radiolucency in the right hemimandible, with the lower border of the mandibular cortical bone extending inferiorly (Figure 2). Computed tomography (CT) revealed a bone-resorption lesion with well-defined borders in the right mandibular body involving the roots of

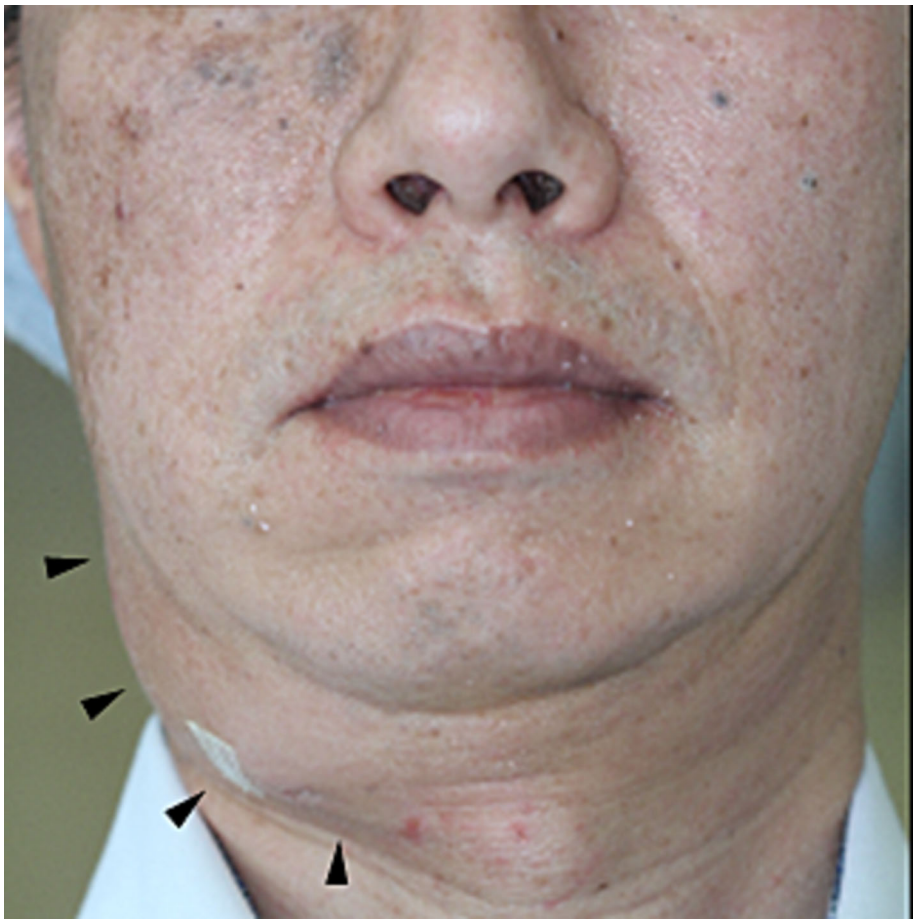

FIGURE 1 Facial photograph at the first medical examination. Black arrowheads indicate swelling in the right submandibular region.

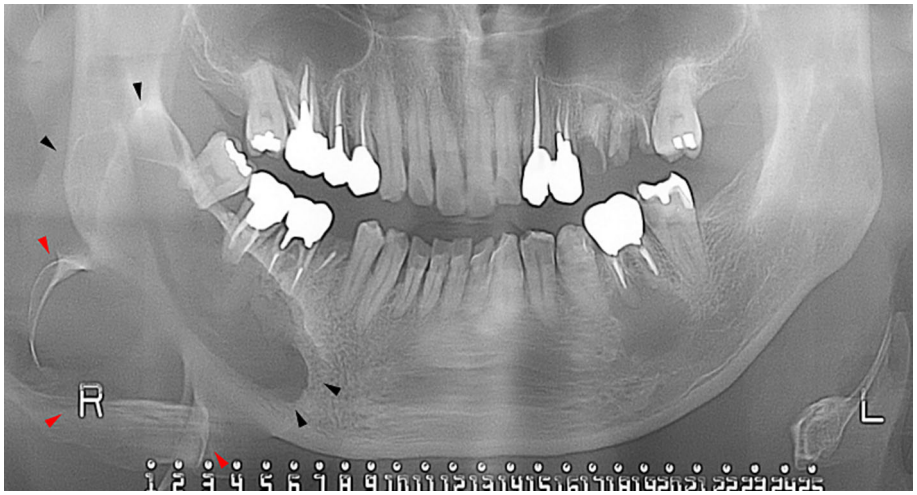

FIGURE 2 Panoramic radiograph at the first examination. A huge well-defined oval radiolucency is observed in the right posterior mandible (black arrowheads), and the bone extends caudally (red arrowheads).

the mandibular right first, second, and third molars. Although mild root resorption was observed in the mandibular third molar, it was not evident on the first or second molars. The lesion exhibited a marked buccolingual bulge. The cortical bone on the lingual side had largely eroded, and the suprahyoid muscles and hyoid bone were displaced to the left side, leading to airway constriction. Moreover, the lesion extended to the submandibular region, beyond the lower border of the mandible, with eggshell-like thickened bone tissue on the buccal side. The margin of the newly formed bone was lobulated (Figures 3 and 4).

We performed a biopsy and fenestration of the cystic lesion, along with the extraction of the mandibular right first, second, and

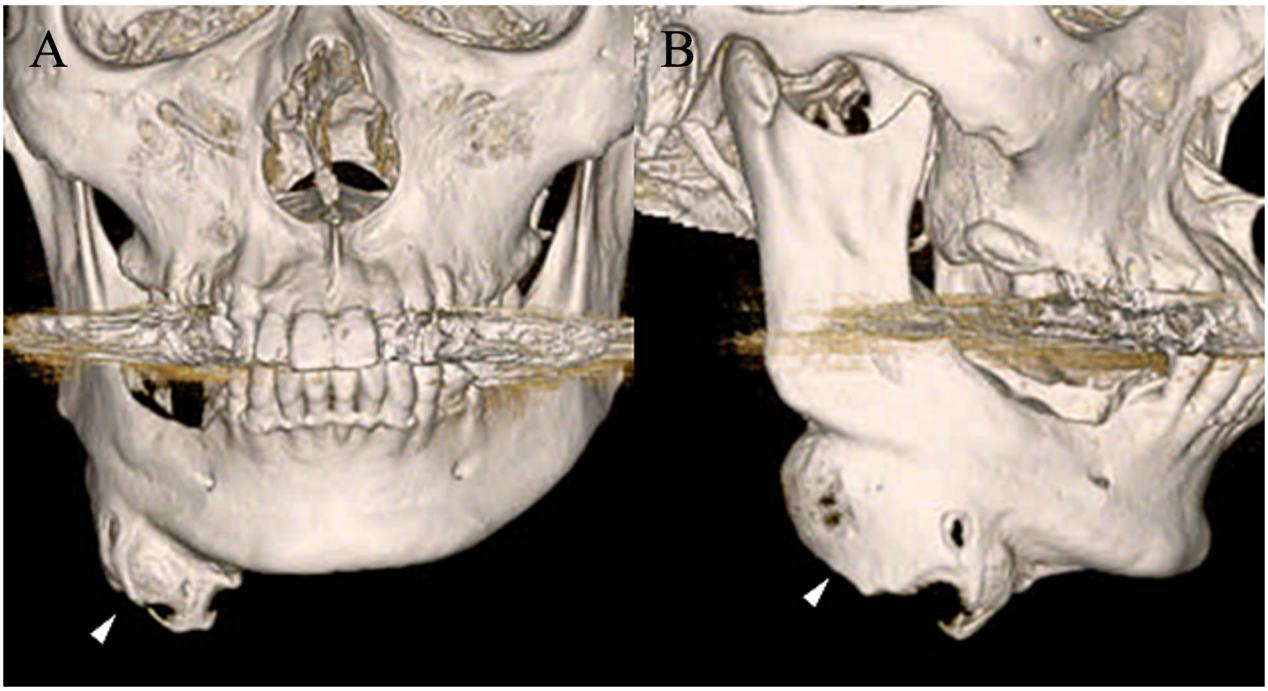

FIGURE 3 Three-dimensionally reconstructed computed tomography images. A continuous inferior border of the mandible extending caudally is observed (white arrowhead). (A) Frontal image; (B) lateral image.

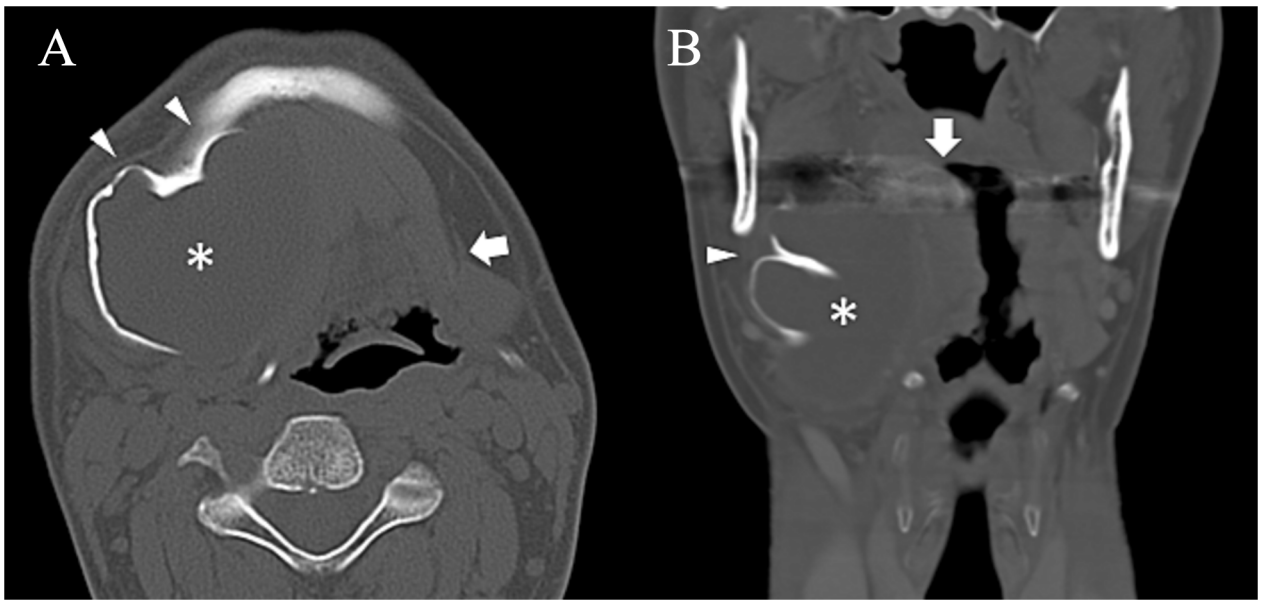

FIGURE 4 Preoperative computed tomography images. A well-defined low-density area is observed in the right mandible (asterisk) with a marked buccolingual bulge and shell-like thickened lobulated bone tissue on the buccal side (white arrowhead). The cortical bone on the lingual side has largely eroded, and the suprahyoid muscles are displaced to the left side (white arrow). (A) axial slice; (B) coronal slice.

third molars under local anesthesia. A small amount of muddy contents was observed in the lesion. Histopathological examination of the specimen revealed a two-layered cyst lining composed of a non-keratinized stratified squamous epithelial covered by fibroconnective tissue. In addition, inflammatory cell infiltration was observed in the subepithelial tissue of the cyst. The histopathological findings suggested a radicular cyst, classified as an odontogenic cyst of inflammatory origin, and a secondarily infected odontogenic developmental

cyst as differential diagnoses; however, distinguishing between them through the biopsy was challenging. During the 8 month fenestration period, the lesion size reduced from 65 mm (long diameter) × 46 mm (short diameter) × 78 mm (craniocaudal diameter) to 58 × 25 × 45 mm. Thereafter, we performed percutaneous cyst enucleation and osteoplasty of the lower border of the mandible to inspect the pathology of the bone extending into the submandibular aria and to improve the esthetic appearance of the jaw, under general anesthesia (Figure 5). Considering that the histopathological features of the resected tissue specimen were similar to those at the time of biopsy, we finally diagnosed this case as an odontogenic cyst, Not Otherwise Specified (NOS). The resected bone tissue was fibrous in nature (Figure 6). Postoperative healing was uneventful, including the restoration of mandibular bone morphology (Figure 7); no recurrent lesions were observed in the mandible 2 years postoperatively. The patient's face displayed bilateral symmetry, and panoramic radiography (Figure 8) and CT images revealed the formation of mandibular cortical bone (Figure 9).

3 | DISCUSSION

This case highlights the atypical progression of odontogenic cyst in the submandibular region, accompanied by the formation of thickened bone. Enlarged odontogenic cysts are generally associated with cortical bone thinning and expansion. Ragsdale et al. describe that any widening of bone contour represents periosteal activity, and general bone cysts can lead to the formation of a circumferential shell, completely replacing the original cortex around the perimeter due to periosteal reaction.[4] Periosteal reactions can be roughly categorized into two types, continuous and interrupted, according to the formation process.[5] The shell-pattern periosteal reaction is a type of continuous periosteal reaction, which is further classified into smooth and lobulated types.[5] It occurs because of the synchronous and continuous inner cortical resorption and outer periosteal formation of new bone, which leads to outward bulging of the cortical bone.[4] While the cortical bone dynamics of jaw bone is morphologically thought to be very similar to the periosteal reaction, proving this pathologically is somewhat challenging, and no existing reports define it as periosteal reaction. Therefore, we decided to recognize it as a periosteal-like reaction. Whether inflammatory or developmental in origin, odontogenic cysts commonly exhibit a thin shell-type cortical bone expansion. However, a few cases of jaw cysts/tumors with thickened shell-pattern bone, including odontogenic keratocyst, unicystic ameloblastoma, aneurysmal bone cyst, and ameloblastic carcinoma, have been reported in the literature.[6–9] In the present case, thickened lobulated shell-pattern bone formation was observed outside an odontogenic cyst, which has not been previously documented. Pathological examination revealed infiltration of inflammatory cells into the partial subepithelial tissue of the cyst. The immune system is deeply involved in bone remodeling, and stimulation by inflammatory cytokines and prostaglandins released from immune cells promotes osteogenesis by acting on the osteoblasts and osteoclasts present in the inner cambial layer of the periosteum.[10] Son et al. reported a case of an inflammatory dentigerous cyst showing increased uptake in the peripheral rim on bone scan.[11] Gupta et al. reported a case of an inflamed unicystic ameloblastoma forming thickened shell-pattern bone tissue, suggesting that

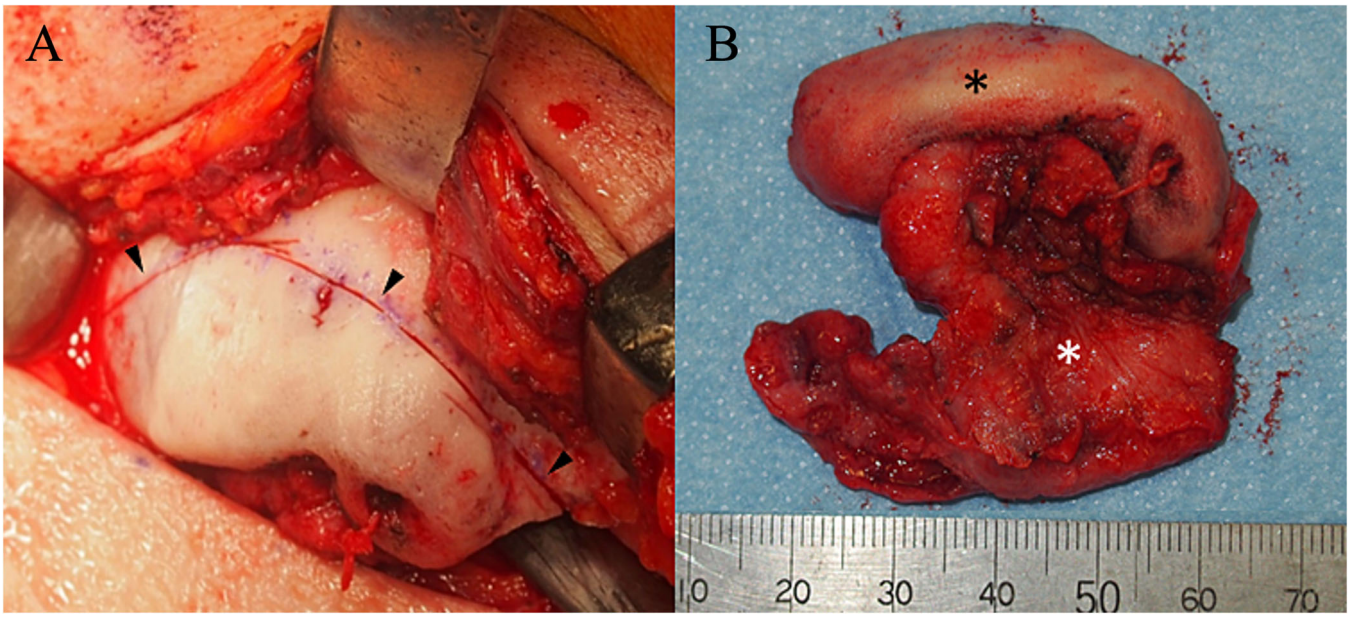

FIGURE 5 (A) Intraoperative photograph showing shell-like bone tissue. The arrowheads indicate the osteotomy line. (B) The extracted specimen is shown. Black and white asterisks indicate the bone tissue and cyst, respectively.

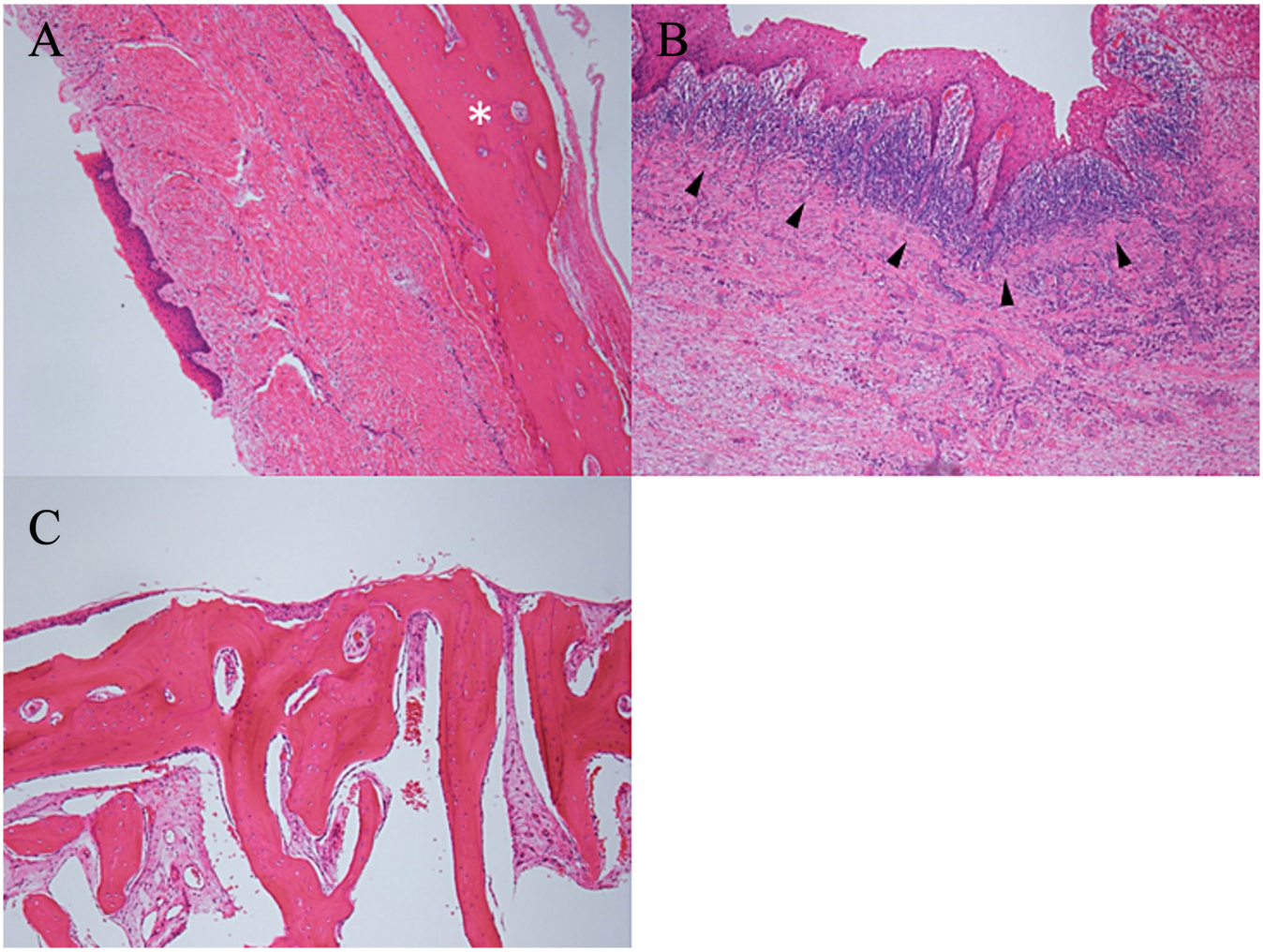

FIGURE 6 Hematoxylin and eosin stained histological specimen. (A) The cyst lining is composed of a non-keratinized stratified squamous epithelial covered by fibroconnective tissue. * shows periosteum. Magnification, 100×. (B) Infiltration of inflammatory cells is observed under the cystic epithelium (black arrowhead). Magnification, 100×. (C) View of the inner surface of the formed bone. Due to rapid bone resorption, the bone remained within the cyst cavity was spiculated. Magnification, 200×.

inflammation promotes osteogenesis around jaw cysts.[6] Formation of shell-pattern cortical bone requires a sufficient duration.[4,5] Thus, in this case, the prolonged existence of extensive cystic lesion and inflammatory stimulation associated with secondary infection could have possibly induced the formation of a thickened shell-type bone through a periosteal-like reaction.

Pathologically distinguishing between an odontogenic cyst of inflammatory origin and a secondarily infected odontogenic developmental cyst, as previously reported,[12] proved challenging in this case. However, the presence of a lobulated shell pattern along the bone margin, attributed to the periosteal-like reaction, led to the suspicion that this case was not a unilocular lesion but a multilocular lesion. Thus, it was inferred that this case was possibly not an inflammatory odontogenic cyst that is usually unilocular but a developing odontogenic cyst based on the multilocular appearance.[13] Although the periosteal-like reaction is not specific, it reflects the characteristics of the underlying lesion and is an important point for differential diagnosis; therefore, it was suggested that the periosteal-like reaction could contribute to the diagnosis of odontogenic cysts.

Mandibular odontogenic cysts extending into the submandibular region can potentially lead to airway stenosis, necessitating urgent airway management. Upper-airway stenosis is more likely to occur in cases with complete resorption of the lingual cortical bone, as lesion expansion medially becomes easier. In the present case, the cyst was enlarged, extending into the submandibular region with bone formation, leading to upper airway constriction as the hyoid bone and suprahyoid muscles were markedly displaced to the contralateral side. Further medial expansion caused by acute inflammation due to infection could have led to severe airway constriction. In addition, the thickened bone tissue on the buccal side of the submandibular region might have contributed to airway constriction. Therefore, early treatment was initiated.

Several aspects concerning the mechanism by which the present case manifested its dynamics remain unclear. Factors other than

FIGURE 7 Facial photograph after 2 years of surgery. The mandibular bone is symmetrical, and no swelling is observed.

FIGURE 8 Panoramic radiograph after 2 years of surgery. No findings suggestive of recurrence are observed in the mandible, and bone healing appears to be normal.

duration and inflammation should be examined. In addition, the lobular bone morphology observed in this case did not necessarily indicate the morphology of the lesion, as jaw cysts are strongly influenced by the environment surrounding the lesion or the original anatomical structure of the jaw. Moreover, a few cases of radicular cysts and odontogenic cysts of inflammatory origin showing multilocular radiolucency have been reported,[14,15] leaving open the possibility that the lesion in this case could be an inflammatory odontogenic cyst.

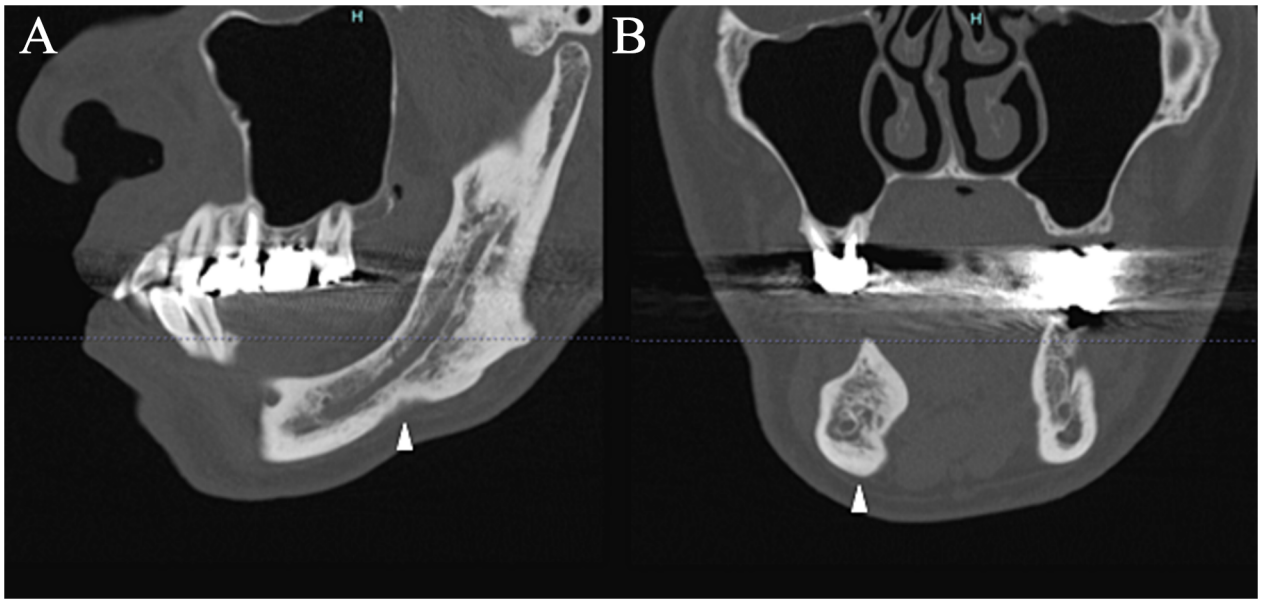

FIGURE 9 Computed tomography images at 2 years postoperatively. Formation of normal mandibular cortical bone is observed (white arrowhead. (A) sagittal slice; (B) coronal slice.

In conclusion, we propose that odontogenic cysts can form a thickened shell-pattern bone tissue because of a periosteal-like reaction, and the morphology of the formed bone is useful for diagnosis. Most importantly, early examination of enlarged odontogenic cysts extending into the submandibular region is necessary because of the possibility of latent airway stenosis.

AUTHOR CONTRIBUTIONS

Shingo Kodama contributed to surgery, postoperative management, conceptualization, data curation, data analysis, and writing. Toshihiro Uchihashi contributed to patient's examination, determination of the treatment plan, surgery, postoperative management, conceptualization, data curation, data analysis, and writing. Hiroaki Shimamoto contributed to interpretations of radiograms. Toshihiro Inubushi contributed to supervision. Yu Usami contributed to histological analyses. Susumu Tanaka contributed to determination of the treatment plan, surgery, and supervision. All authors reviewed the manuscript.

ACKNOWLEDGMENTS

None.

CONFLICT OF INTEREST STATEMENT

The authors declare no conflicts of interest.

DATA AVAILABILITY STATEMENT

N/A

ETHICS STATEMENT

All the clinico-pathologic investigations detailed in the manuscript have been conducted in accordance with the Declaration of Helsinki and its later amendments or comparable ethical standards.

PATIENT CONSENT STATEMENT

Written informed consent was obtained from the patient for publication of this case report and accompanying images.

CLINICAL TRIAL REGISTRATION

N/A

ANIMAL STUDIES

N/A

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How to cite this article: Kodama S, Uchihashi T, Shimamoto H, Inubushi T, Usami Y, Tanaka S. Mandibular odontogenic cyst atypically extended into the submandibular region with thickened bone formation: A case report. Oral Sci Int. 2024. https://doi.org/10.1002/osi2.1228