

# Primary xanthoma inferior to the right mandibular third molar and intraoral vertical ramus osteotomy

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## ABSTRACT

Intraosseous xanthoma of the mandible is a rare benign disorder. A 17-year-old male patient presented with a suspected abscess in the right mandibular third molar, detected on a panoramic radiograph. The patient had no history of systemic or lipid-related metabolic diseases and complained of no specific symptoms or pain. A radiographic examination revealed a heterogeneous radiolucency extending from the apical to the distal aspect of the right mandibular third molar tooth germ. The lesion measured 9 × 16 × 24 mm (antero-posterior × medio-lateral × supero-inferior) and showed a relatively well-defined, multilocular, foamy appearance with hyperostotic borders spreading to the inferior alveolar nerve canal. After excisional biopsy, a diagnosis of central xanthoma was made. The lesion recurred, and intraoral vertical ramus osteotomy was done near the lesion. For the treatment of xanthoma of the mandible, extensive and delicate surgical treatment under general anesthesia should be considered. (*Imaging Sci Dent* 2022; 52: 231-8)

**KEY WORDS:** Xanthoma; Mandible; Orthognathic Surgery; Mandibular Osteotomy

Xanthoma is a benign soft tissue lesion that primarily occurs in the skin after minor trauma or friction.<sup>1</sup> The term “xanthoma” is derived from the Greek word *xanthos*, which means “yellow,” because changes in lipid metabolism lead to yellow pigment deposition in the skin and other organs.<sup>2,3</sup> Xanthoma occasionally occurs in association with metabolic diseases, such as type II or III hyperlipidemia and diabetes.<sup>4,5</sup>

Xanthoma occurring in the bone of patients with no history of systemic or lipid-related metabolic diseases is called primary xanthoma.<sup>6</sup> Rawal et al.<sup>7</sup> reviewed cases of primary xanthoma that were not associated with other bone or soft tissue lesions or with a history of hyperlipidemia. They presented a list of 8 features that distinguish central xanthoma of the jaw bones from other conditions.

Primary xanthoma occurs rarely, and it presents as a lytic, expansile lesion on radiographic images.<sup>6</sup> To date, no studies have reported further surgical treatment, such as orthog-

nathic surgery of the mandible, after excision of a mandibular xanthoma. Herein, the current report presents a rare case of mandibular primary xanthoma for which orthognathic surgery of the mandible was performed following excision of a recurrent lesion.

## Case Report

The study was approved by the Institutional Review Board of National Health Insurance Service Ilsan Hospital (No. NHIMC 2021-11-006), and the requirement for informed consent was waived by the IRB.

A 17-year-old male patient presented at the Department of Oral and Maxillofacial Surgery at the National Health Insurance Service Ilsan Hospital with a suspected abscess in the area of the right mandibular third molar, detected on a panoramic radiograph. The patient had no history of systemic or lipid-related metabolic diseases. The patient complained of no specific symptoms or pain, and a clinical examination revealed no abnormal findings in the soft tissue around the right mandibular third molar. A panoramic radiograph and cone-beam computed tomography (CBCT) images were acquired at the first visit. The radiographic

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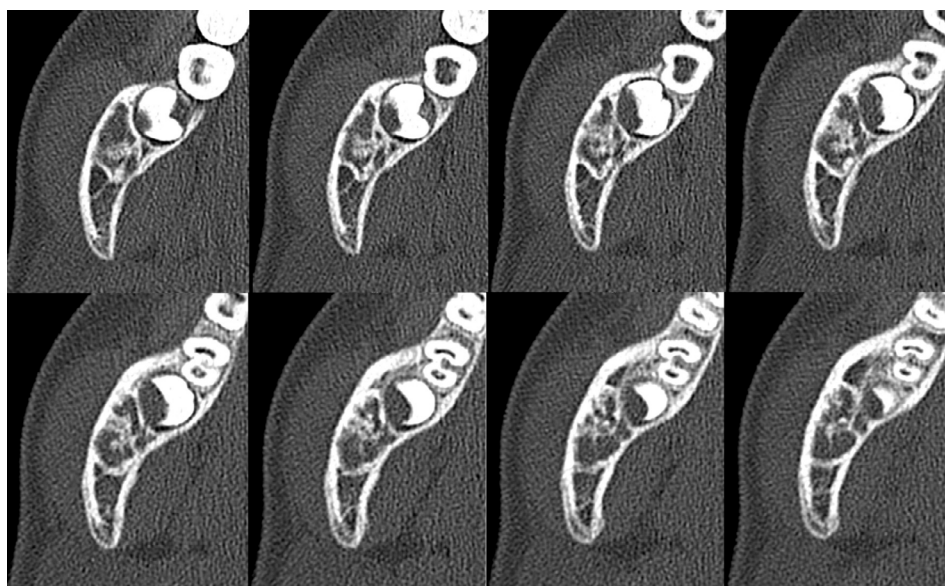
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**Fig. 1.** Panoramic radiography showed a heterogeneous radiolucency with a soap bubble appearance extending from the apical to distal areas of the right mandibular third molar tooth germ at the first examination.



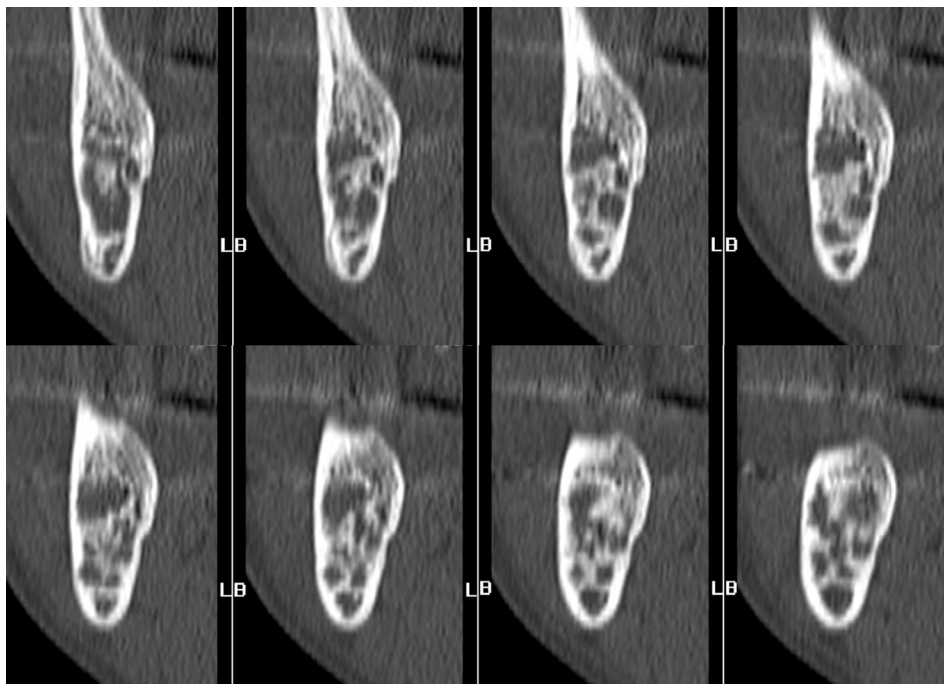
**Fig. 2.** Serial axial images of cone-beam computed tomography at the first examination. Images show no bony bulging and periosteal reaction near the right mandibular lesion. The lesion shows a relatively well-defined, multilocular, foamy appearance with hyperostotic borders and shows a mixed pattern of radiolucent and opaque regions. The lesion is near the apex of the right mandibular third molar, but the border of the lesion is separated. The trabecular bone in the posterior mandible is replaced by radiolucency and opacity. The size of the lesion is about 9 mm × 16 mm (antero-posterior × medio-lateral).

examination revealed bilateral impacted mandibular third molars and a heterogeneous radiolucency extending from the apical to distal areas of the right mandibular third molar tooth germ in the panoramic view (Fig. 1). In the CBCT images, the lesion measured 9 mm × 16 mm × 24 mm (antero-posterior × medio-lateral × supero-inferior) and showed a relatively well-defined, multilocular, foamy appearance with hyperostotic borders spreading to the inferior alveolar nerve canal (Figs. 2 and 3).

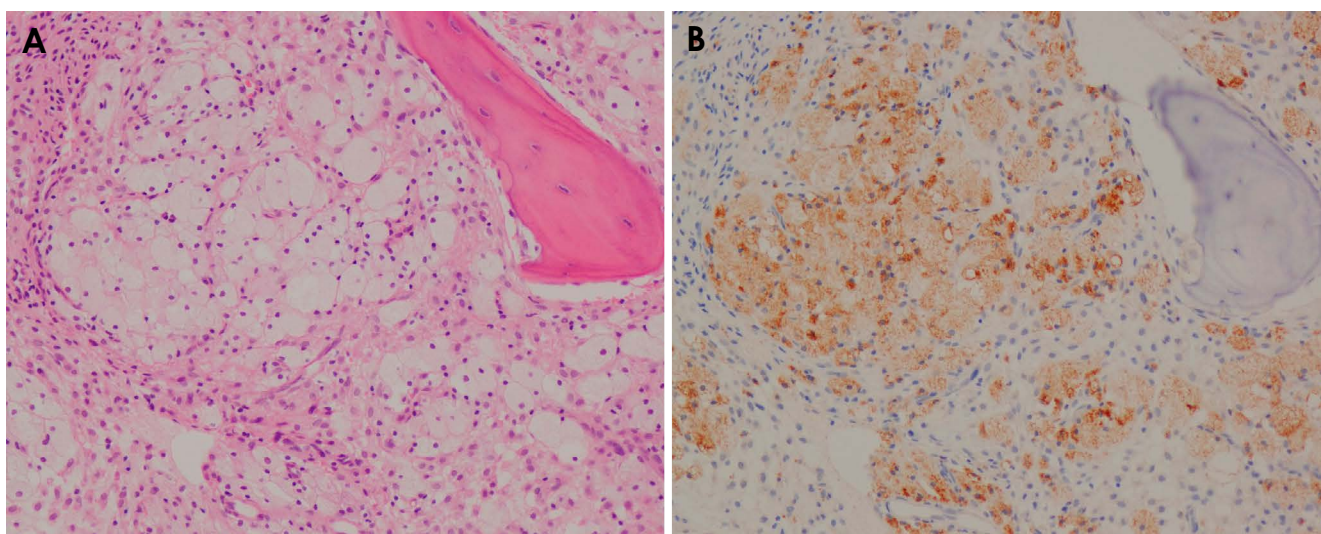
An excisional biopsy was performed, and the right mandibular third molar was extracted using osteotomy and odontotomy under local anesthesia at the patient's next visit. The lesion was not a single cohesive lump, and it had a mushy consistency. The lesion was clearly removed by curettage

from the surrounding tissue, and part of the specimen was sent to the Department of Pathology. A diagnosis of central xanthoma was made, and the sutures were removed 1 week after surgery. Sheets of foamy and non-foamy macrophages were observed under a microscope, and immunohistochemical staining showed positivity for CD68 (Fig. 4).

Since many cases of xanthoma are associated with systemic lipid metabolism disorders, a complete blood count was performed, including cholesterol, triglyceride, and high- and low-density lipoprotein cholesterol levels. The patient showed a normal lipid profile. At a follow-up examination, a panoramic radiograph taken 5 months after surgery revealed a radiolucency in the right posterior mandible, with a similar pattern to that observed at the first visit.



**Fig. 3.** Serial sagittal images of cone-beam computed tomography at the first examination. Instead of the normal structure of trabecular bone, it is observed as a bubble or honeycomb appearance. The size of the lesion is about  $16 \times 24$  mm (medio-lateral  $\times$  supero-inferior). A particularly radiopaque mass is observed inside the lesion with hyperostotic border and a radiolucent rim around it. The lesion is near the right inferior alveolar nerve. Displacement of the inferior alveolar nerve is not observed.



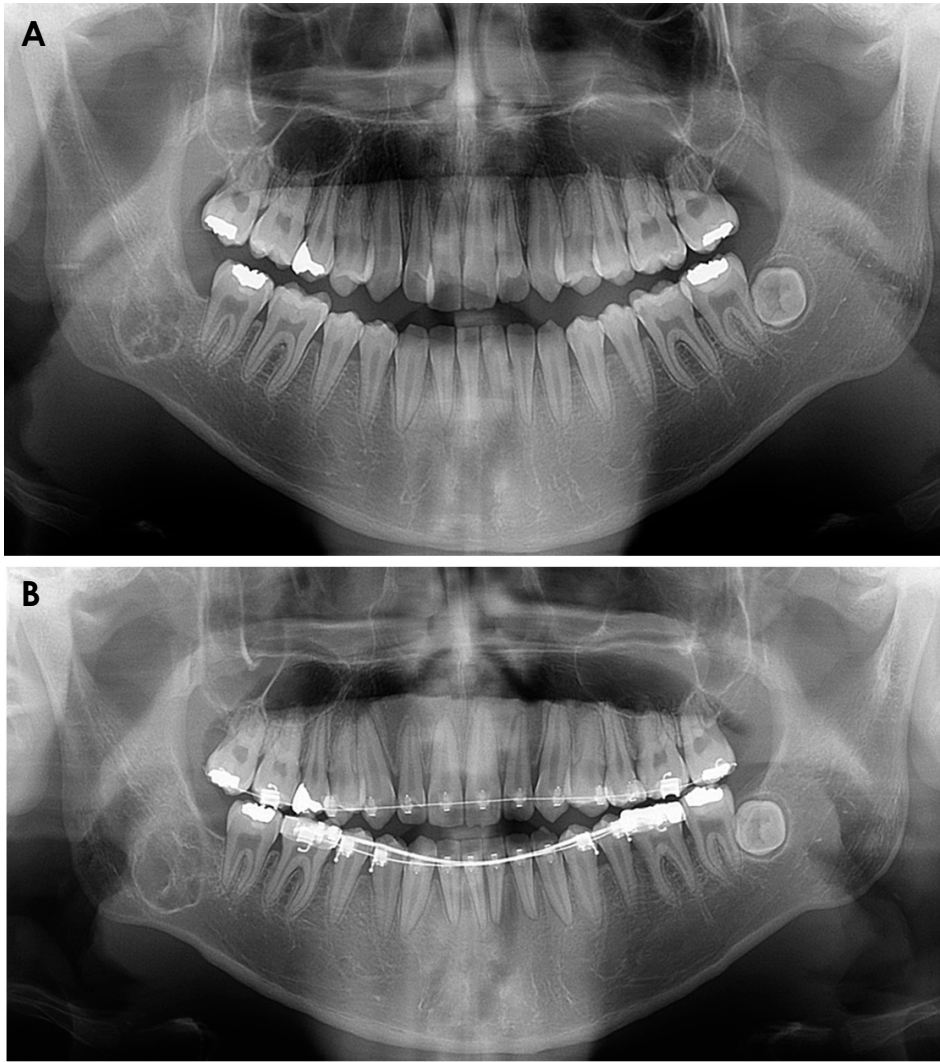
**Fig. 4.** A. The microscopic features of the lesion comprise sheets of lipid-laden foamy histiocytes with presence of bone fragments and mature adipocytes (hematoxylin and eosin stain, original magnification  $\times 20$ ). B. Immunohistochemical properties of the lesion. The xanthoma cells are strongly and diffusely positive for CD68 (original magnification  $\times 20$ ).

A panoramic radiograph taken 1 year and 8 months after surgery showed enlargement of the lesion (Fig. 5A and B). On a multidetector computed tomography examination for the reoperation taken with panoramic images, the lesion showed a honeycomb-like structure with a septum and a hyperostotic border. Compared with the previous CBCT scan, internal radiopacity was observed broadly throughout the lesion. Similar to the previous CBCT scan, the contrast of the intra-lesion radiolucency was similar to that of the

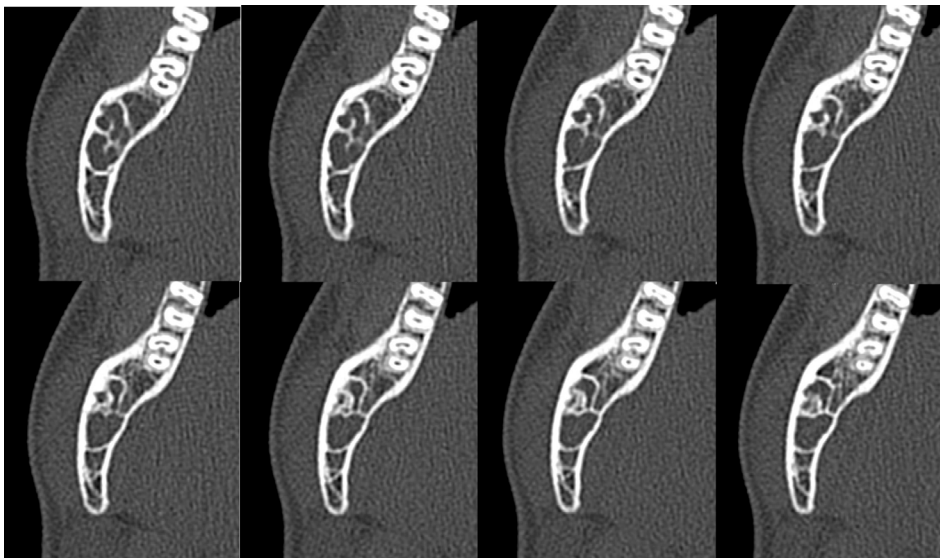
adjacent soft tissue (Figs. 6 and 7).

Subsequently, the mass was removed by curettage under general anesthesia (Fig. 8). No specific findings were found other than slight paresthesia in the right lower lip; a diagnosis of primary bone xanthoma was rendered, as in the first operation. At a 1-month follow-up after reoperation, healing was normal, and the patient re-visited the hospital 2 months after reoperation to receive orthognathic surgery.

The patient received presurgical orthodontic treatment

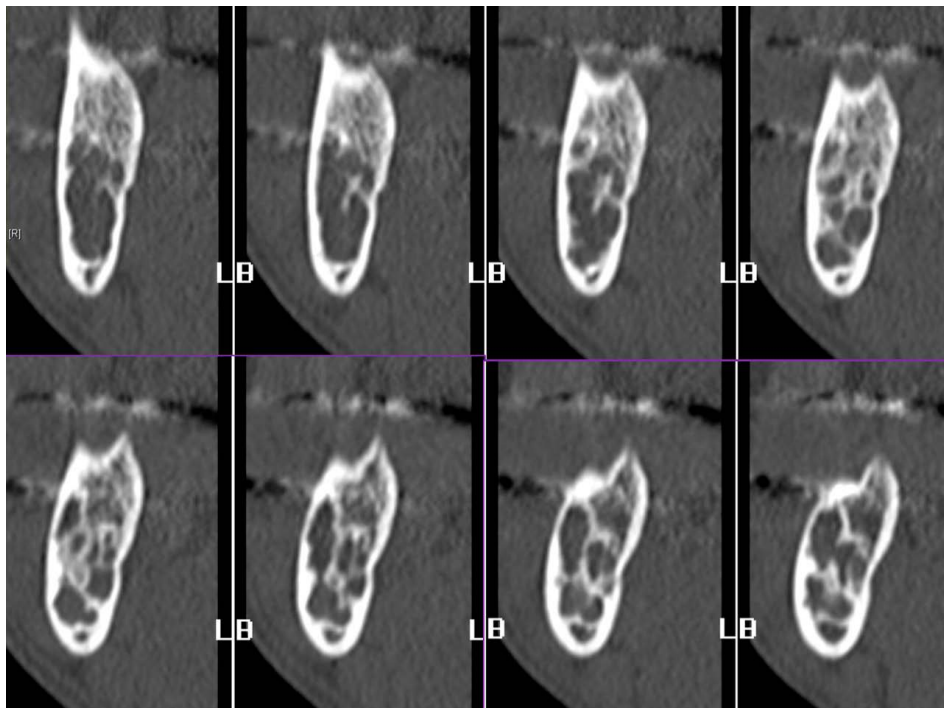


**Fig. 5.** A. A panoramic radiograph taken 5 months after surgery, revealing enlargement of the lesion and similar findings to those observed at the first visit. B. A panoramic radiograph taken 1 year and 8 months after surgery, showing a larger lesion than at 5 months after surgery.

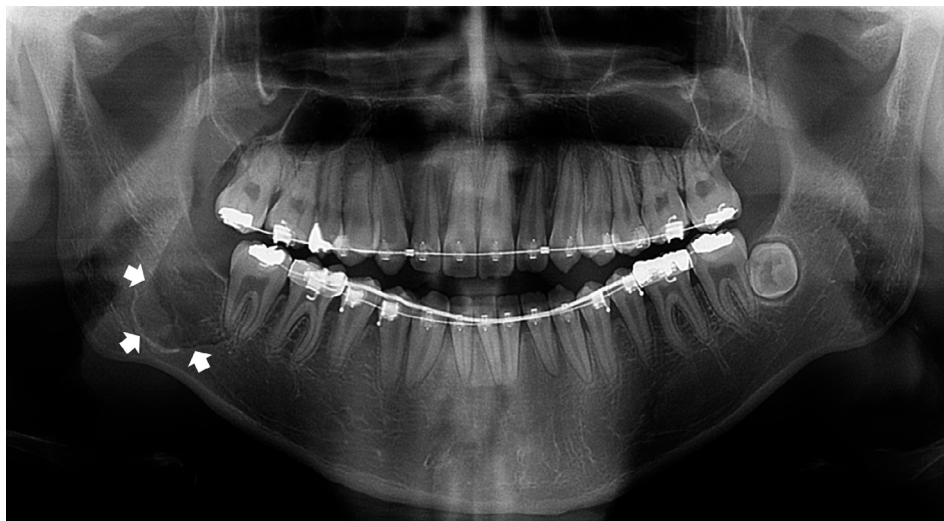


**Fig. 6.** In serial axial images of a multidetector computed tomography scan for the reoperation, lesion shows honeycomb-like structure with septum, hyperostotic border. Compared with the previous cone-beam computed tomography (CBCT) scan, broad internal radiopacity is observed throughout the lesion. Similar to the previous CBCT scan, the contrast of the intra-lesion radiolucency is similar to that of the adjacent soft tissue. Compared with the previous CBCT scan, the size of the radiopaque mass inside the lesion is smaller, and the honeycomb-shaped structure and septum are more clearly observed.





**Fig. 7.** In serial sagittal images of a multidetector computed tomography scan compared with the previous cone-beam computed tomography (CBCT) scan, the septum inside the lesion with the hyperostotic border is more distinct. Similar to the previous CBCT scan, a honeycomb-shaped structure with a septum inside the lesion is observed, similar to the polymorphic structure, but the spaces in the lesion are smaller and denser than in the previous CBCT scan. Thinning and bulging of the mandible to the inferior border are found. The radiopaque mass inside the lesion is small, and it is observed as a large number of spaces with polymorphic structures. Various degrees of contrast are observed for each.

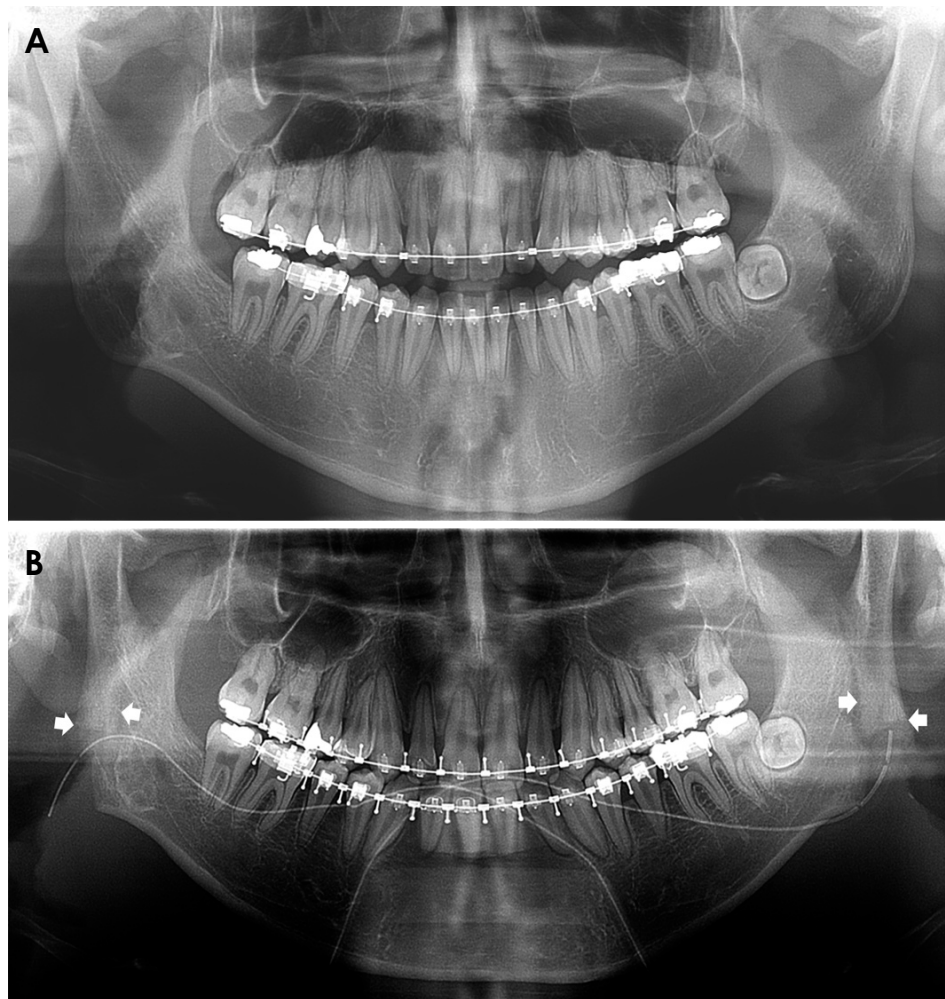


**Fig. 8.** A panoramic radiograph taken immediately after surgery under general anesthesia, revealing wide excision of the lesion. White arrows indicate the surgical site.

for mandibular prognathism, which was started after the xanthoma removal. At the time of orthognathic surgery, the alignment and leveling of the teeth were ready. However, since orthognathic surgery was performed in the mandibular ramus, the timing and methods of surgery were carefully determined, considering the possibility of recurrence and spread. In addition, since the bone defect in the ascending ramus had to be repaired before orthognathic surgery, the authors decided to perform orthognathic surgery after further follow-up visits.

As considerable bone formation was confirmed on pano-

ramic views at 4- and 7-month follow-up visits after the reoperation, orthognathic surgery was performed (Fig. 9). The mandible could be set back using sagittal split ramus osteotomy (SSRO) or intraoral vertical ramus osteotomy (IVRO). However, SSRO would have included the previous xanthoma region; therefore, IVRO was selected as the surgical method. Bilateral IVRO was performed in consultation with the Department of Orthodontics, and postoperative recovery was uneventful (Fig. 9B). After 2 weeks of intermaxillary fixation, mouth-opening exercises were carried out for approximately 3 weeks. The patient showed



**Fig. 9.** A. A panoramic radiograph taken 7 months after surgery, revealing no recurrence of the lesion. B. A panoramic radiograph taken immediately after intraoral vertical ramus osteotomy (IVRO). IVRO was performed 9 months after xanthoma removal. White arrows indicate the ramus osteotomy site. The white lines are Jackson-Pratt suction tubes.



**Fig. 10.** A panoramic radiograph taken at 20 months (29 months after the removal of xanthoma) revealed no recurrence of the lesion after intraoral vertical ramus osteotomy.

a favorable range of mouth opening, as well as anterior and lateral movements.

Although a slightly atypical lesion was observed in the right mandibular angle on a 7-month follow-up panoramic

radiograph, no signs of recurrence were observed on radiographic examinations for approximately 2 additional years, and the areas of orthognathic surgery had healed (Figs. 9 and 10).

## Discussion

Since Slootweg et al.<sup>8</sup> reported a xanthomatous lesion of the mandible in 1993, several cases of primary mandibular xanthoma have been reported.<sup>6,8-10</sup> These reports stated that primary xanthoma can be treated using complete removal by curettage with a favorable prognosis, and few studies reported recurrence of the lesion.

The patient described herein was an otherwise healthy 17-year-old male with no underlying conditions, such as hyperlipidemia or diabetes. If there is evidence of an endocrine or metabolic disease leading to elevated cholesterol levels, such as primary hyperlipidemia, hypolipoproteinemia type II and III, diabetes, hypothyroidism, or lipoprotein lipase deficiency, xanthoma of the mandible is called secondary bone xanthoma.<sup>5,11,12</sup> At the first visit, radiographic images, including CBCT images, did not show bulging and thinning of the mandibular cortex or reactive periosteal bone formation. A heterogeneous lesion and soap bubble appearance were found on the radiograph. The patient was diagnosed with primary xanthoma based on excisional biopsy by curettage.

The lesion likely recurred because it had been incompletely excised during the first surgery, or because it was not well-encapsulated as a solid mass, precluding complete excision by curettage alone. For this reason, the recurrent lesion was removed by curettage with peripheral ostectomy, which may have led to complete excision with no further recurrence. Surgeons removing xanthomas should take this into account. An excellent prognosis following surgery by curettage was reported.<sup>13</sup>

As in the present case, xanthoma located in the area of forthcoming orthognathic surgery should be treated using osteotomy techniques, such as IVRO or mandibular subcondylar osteotomy, to avoid the lesion site and to prevent recurrence and spread of the lesion. Conversely, SSRO should not be used.

In the present case, histopathological examination showed sheets of foamy and non-foamy macrophages that were positive for CD68 immunohistochemistry. Histopathologically, histiocyte-related diseases can be divided into Langerhans cell histiocytosis and non-Langerhans histiocytic processes.<sup>12</sup> Xanthoma belongs to the latter category and shows a prominent accumulation of lipid-laden macrophages or foam cells with multinucleated giant cells. Spindle cells are occasionally found; this is a distinct feature of benign fibrous histiocyte of bone, which can be easily confused with central xanthoma.<sup>13-15</sup>

It is difficult to diagnose xanthoma by imaging findings.<sup>1</sup>

In mandibular xanthoma, the radiographic appearance varies widely, from lesions with well-defined sclerotic margins to lesions with diffuse and ill-defined borders.<sup>16,17</sup> In lesions with irregular and ill-defined margins, it is necessary for the differential diagnosis to rule out primary or metastatic malignant lesions of the mandible.

Due to the deposition of calcified material, primary mandibular xanthoma can show diffuse radiologic findings, similar to those of fibrous dysplasia.<sup>3</sup> If osseous xanthoma presents as a well-defined lesion with a sclerotic margin or a small region of sclerosis, it is difficult to differentiate xanthoma from common benign bone lesions, including non-ossifying fibroma, benign fibrous histiocytoma, fibrous dysplasia, simple bone cyst, giant cell tumor, or brown tumor of hyperparathyroidism.<sup>17</sup> Xanthoma can occur in a pre-existing lesion, including a simple bone cyst, aneurysmal bone cyst, and fibrous dysplasia; therefore, careful differentiation of complex lesions is necessary.<sup>4</sup>

The internal appearance of xanthoma varies from a completely radiolucent lesion to a mixed honeycomb lesion, and the peripheral appearance is very diverse, with presentations including corticated, scalloped, ill-defined, and slightly sclerotic margins.<sup>9</sup> Root resorption and inferior alveolar nerve canal displacement are sometimes seen, but xanthoma does not seem to significantly affect the surrounding anatomical structures in the mandible.<sup>7</sup> The rarity of the lesion, its non-specific radiological characteristics, and its atypical radiological appearance make it difficult to diagnose xanthoma by imaging findings.<sup>1</sup>

Scintigraphy also shows non-specifically increased uptake, and a preoperative incisional biopsy may be helpful for diagnosis, but it is insufficient to make an accurate diagnosis in some cases of xanthoma, so complete removal and histopathological examination results are required to diagnose a lesion as xanthoma.<sup>1,6,7</sup>

Considering that the present case of primary xanthoma recurred, although xanthoma has a low recurrence rate, there is a need for an in-depth discussion regarding surgical methods to prevent recurrence. Though 1 week after the reoperation, no specific findings were found other than slight paresthesia in the right lower lip, and the patient showed a good prognosis without paresthesia during the follow-up. Further studies on the histological features related to recurrence are needed, and the present case showed that orthognathic surgery can be performed successfully in jaws with a rare lesion. This suggests that the procedure can be used to treat primary xanthoma in the jaw. However, additional case reports are needed to allow generalization. Moreover, since an orthognathic technique that involved bypass was

performed in the present case, further evaluation of other techniques that directly pass through the lesion site is warranted.

The rarity of xanthoma, its non-specific radiological characteristics, and its atypical radiological appearance make it difficult to diagnose xanthoma by imaging findings. Establishing more criteria for evaluating recurrence, the follow-up interval, and the follow-up duration would help delineate more appropriate treatment methods than excision, as a surgical field of view could be secured through sufficient osteotomy under general anesthesia in patients with xanthoma to prevent recurrence. In particular, delicate and careful surgery around the inferior alveolar nerve area is needed to prevent complications, considering that xanthoma does not seem to significantly affect the surrounding anatomical structures in the mandible.

**Conflicts of Interest:** None

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