



Xanthogranulomatous inflammation of the lower jaw bone: a rare case report

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Abstract (J Korean Assoc Oral Maxillofac Surg 2023;49:360-364)

Xanthogranulomatous inflammation (XGI) is an uncommon type of chronic inflammation and is histologically characterized by foamy histiocytes and giant cells. The most common sites of occurrence are kidneys and gallbladder. The etiology remains controversial. Involvement of the lower jaw bone is rare. In this study, we report a case of XGI presenting in the lower jaw.

Key words: Xanthogranulomatous inflammation, Foamy histiocytes, Lower jaw bone

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I. Introduction

Xanthogranulomatous inflammation (XGI) is an uncommon type of chronic inflammation histologically characterized by foamy histiocytes and giant cells. The most common sites of occurrence are the kidneys and gallbladder¹. The etiology remains controversial, and involvement of the lower jawbone is rare. In this study, we report a case of XGI presenting in the lower jaw. Bone involvement of XGI, termed xanthogranulomatous osteomyelitis (XO), often presents as a mass-like lesion extending to adjacent structures, and can mimic infiltrative carcinoma². Central xanthoma can occur in the bone secondary to hyperlipidemia. However, xanthoma may not show suppurative inflammation³. According to Cozzutto and Carbone⁴, xanthoma and foam cells might share a pathogenetic mechanism but have differing etiology.

II. Case Report

1. Patients and methods

An 81-year-old female patient presented to the Department of Oral and Maxillofacial Surgery, Daejeon Sun Hospital, Korea with a chief complaint of pain and gingival swelling on the right body of the mandible. She had undergone extraction of the first premolar in the right mandible 2 years prior. The patient had hypertension, cerebral infarction, and a surgical history of total knee replacement. She had received 2 previous intravenous injections of bisphosphonates (ibandronic acid 3 mg) (13 years prior and in August 2021).

No facial swelling or pus discharge was noted. The patient reported pain and gingival swelling only on the right body of the mandible.(Fig. 1. A, 1. B) A lesion was noted on radiographic (panorama) imaging.(Fig. 1. C; arrow) On the axial computed tomography (CT) scan, a 33.66-mm-sized irregular margined soft tissue mass was detected in the right mandibular body with underlying bone destruction.(Fig. 2. B) A fistula tract was observed along the #44 extracted region.(Fig. 2. A; yellow arrow)

On magnetic resonance imaging (MRI), coronal T2-weighted imaging (repetition time [TR]/echo time [TE], 3,590/69 ms) was performed at slice thickness of 2.5 mm, matrix of 320×320 mm, and 180×180 field of view.(Fig. 2. C) Axial T1-weighted imaging (TR/TE, 770/12 ms) was performed at

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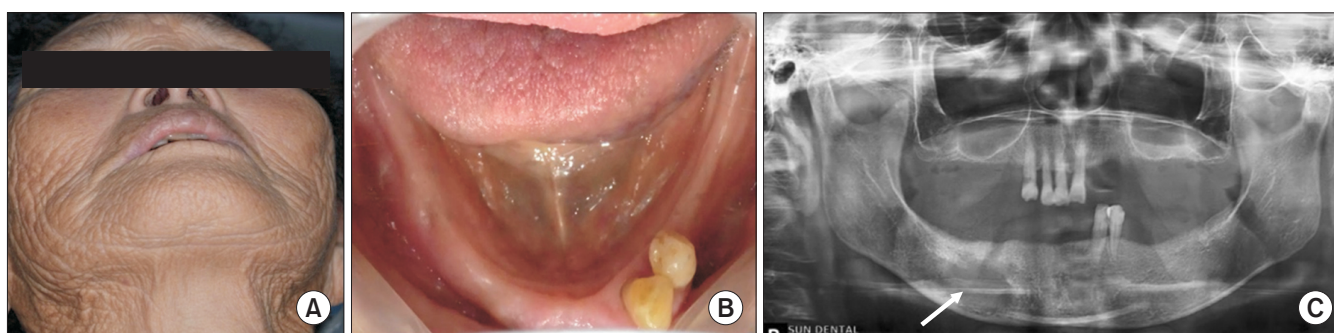


Fig. 1. Preoperative photographs and X-ray. A. No facial swelling and redness on the right body of the mandible. B. Gingival swelling on the right body of the mandible and a fistula tract in the #44 extraction area. C. A radiolucent lesion was observed on X-ray (panorama) imaging (arrow).

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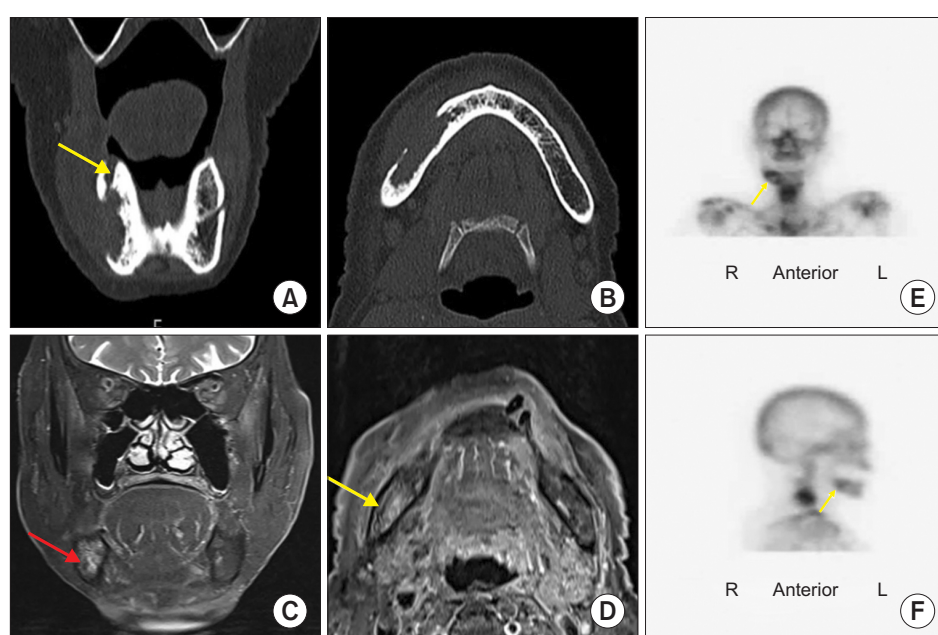


Fig. 2. Preoperative computed tomography (CT), magnetic resonance imaging (MRI), and 3-phase bone scan. A. On CT images, a fistula tract was observed in the #44 extracted region (yellow arrow). B. Bony resorption was noted. C, D. MRI imaging revealed an intraosseous lesion displaying high intensity. The right inferior alveolar nerve (red arrow) was involved in the mass (yellow arrow). E, F. A focal hot spot was observed on the right body of the mandible (yellow arrows).

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slice thickness 3 mm, matrix of 320×272 mm, and a 153×180 field of view.(Fig. 2. C) Signal abnormalities in the mandible were associated with cortical irregularity and diffuse soft tissue hyperintensity.(Fig. 2. C) The right inferior alveolar nerve (IAN) (red arrow) was involved in the mass (yellow arrow). (Fig. 2. C, 2. D) On 3-phase bone scan imaging, faint tracer uptake with right mandible on delayed phasic images.(Fig. 2. E, 2. F; yellow arrows)

Before surgery, incisional biopsy was performed on the #45-#46 bony resorption site to rule out cancer. The biopsy result was chronic inflammation from the Department of Pathology, Daejeon Sun Hospital and XGI from the Department of Pathology, Wonkwang University Daejeon Dental Hospital. Microscopically, abundant foam cells were observed.

Pre-antibiotic (amoxicillin+clavulanate, Newcla 625 mg;

Han Lim Pharm.) treatment and administration of Locfenal 60 mg (loxoprofen sodium hydrate; Han Lim Pharm.) was performed 1 week prior to surgery to reduce inflammation and pain. Surgical excision and neuroplasty were completed under general anesthesia, and a specimen biopsy was performed. Finally, XGI was confirmed by histological sampling.

2. Surgical procedure

Local anesthesia with 2% lidocaine with epinephrine 1:100,000 was administered into the vestibule from the incisor to the third molar of the right mandible. A crestal incision was created, and a mucoperiosteal flap was elevated. A releasing incision around the mental nerve was completed

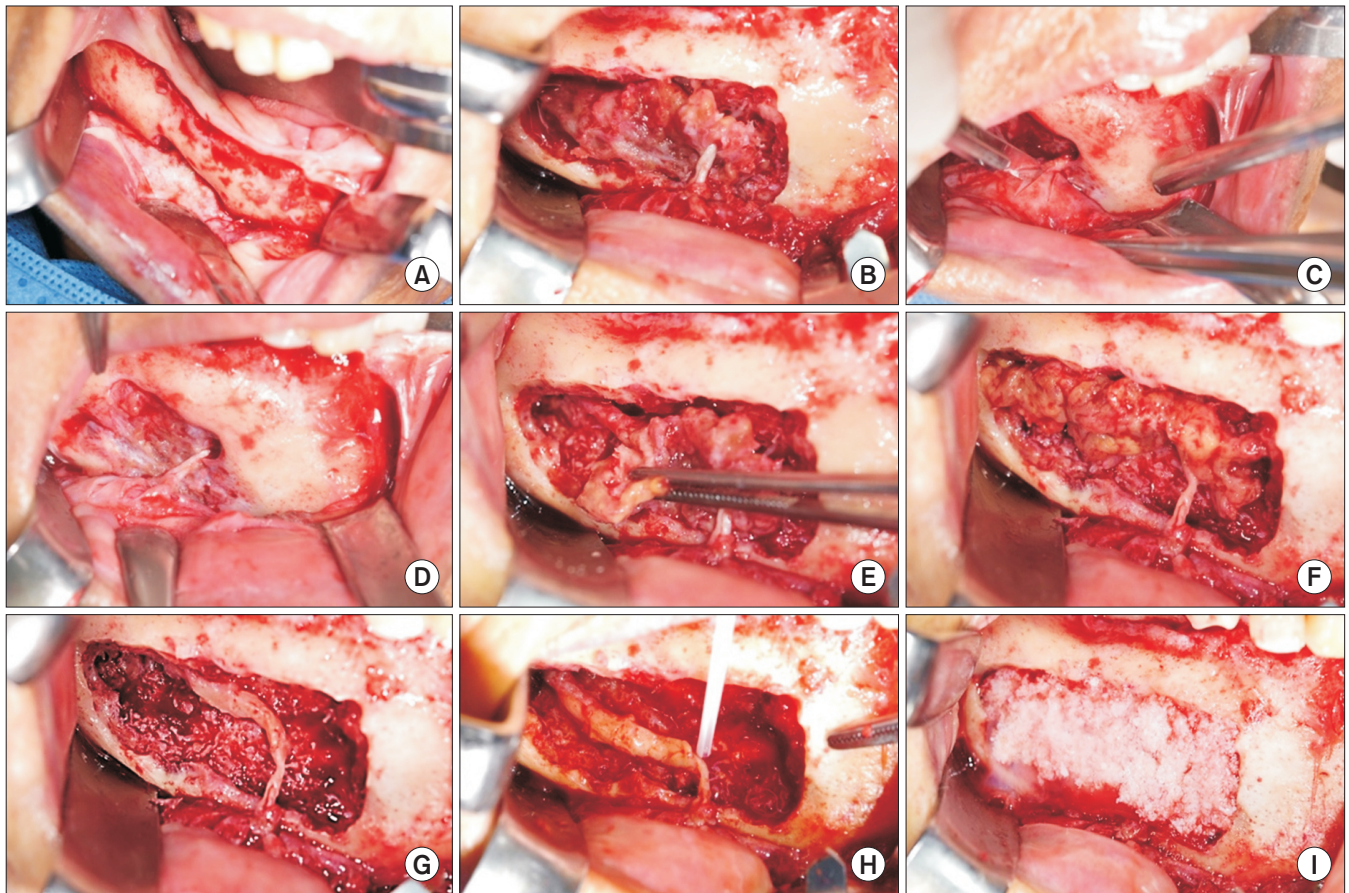


Fig. 3. Surgical procedure. A, B. A crestal incision was created, and a mucoperiosteal flap was elevated to expose the bone and mass. C. A releasing incision around the mental nerve was completed with a #15 blade. D. The mental nerve was dissected from the mass. E. Retrograde dissection of the mental nerve was initiated to identify the inferior alveolar nerve (IAN) anterior to the mass. F. A subsequent anterograde dissection was performed. G. The mass was separated from the IAN. H. The IAN was repositioned to the inferior mandibular border. Guardix-SG (sodium hyaluronate, HA; Hanmi Pharm.), CMC (sodium carboxymethylcellulose; Hanmi Pharm.), and P-STOP (collagen; Ecott Co.) were applied to the IAN to prevent adhesion with the bone graft materials. I. Graft bone (xenograft, bone substitute cancellous granules; Purgo Biologics) material was used to graft the site.

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Fig. 4. The excised mass specimen. A 4-cm-sized, irregularly margined, hard, yellowish mass.

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with a #15 blade.(Fig. 3. C) The tumor was removed while preserving the IAN. Guardix-SG (sodium hyaluronate, HA; Hanmi Pharm.), CMC (sodium carboxymethylcellulose; Hanmi Pharm.), and P-STOP (collagen; Ecott Co.) were applied on the IAN to prevent nerve adhesion to the bone graft materials.(Fig. 3. H) Graft bone (xenograft, bone substitute cancellous granules; Purgo Biologics) material was applied, and the operation site was covered with BioCover (resorbable collagen membrane; Purgo Biologics), an absorbable membrane.(Fig. 3. I)

The specimen was approximately 4 cm in size with irregular margins and presented as a hard, yellowish mass upon gross examination.(Fig. 4)

Upon postoperative microscopic examination, a few necrotic bony fragments with mixed inflammatory cell infiltration consistent with foamy macrophages were observed. Gi-

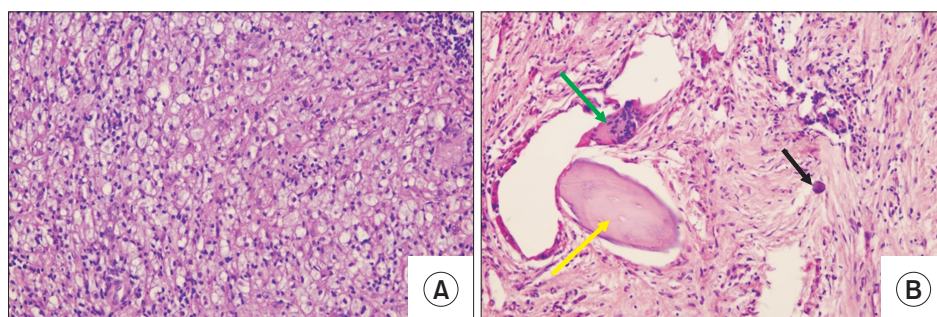


Fig. 5. A. Foam cells. B. Necrotic bone (yellow arrow), histiocytes (green arrow), and fibrosis (black arrow). H&E staining (A, B: $\times 200$).

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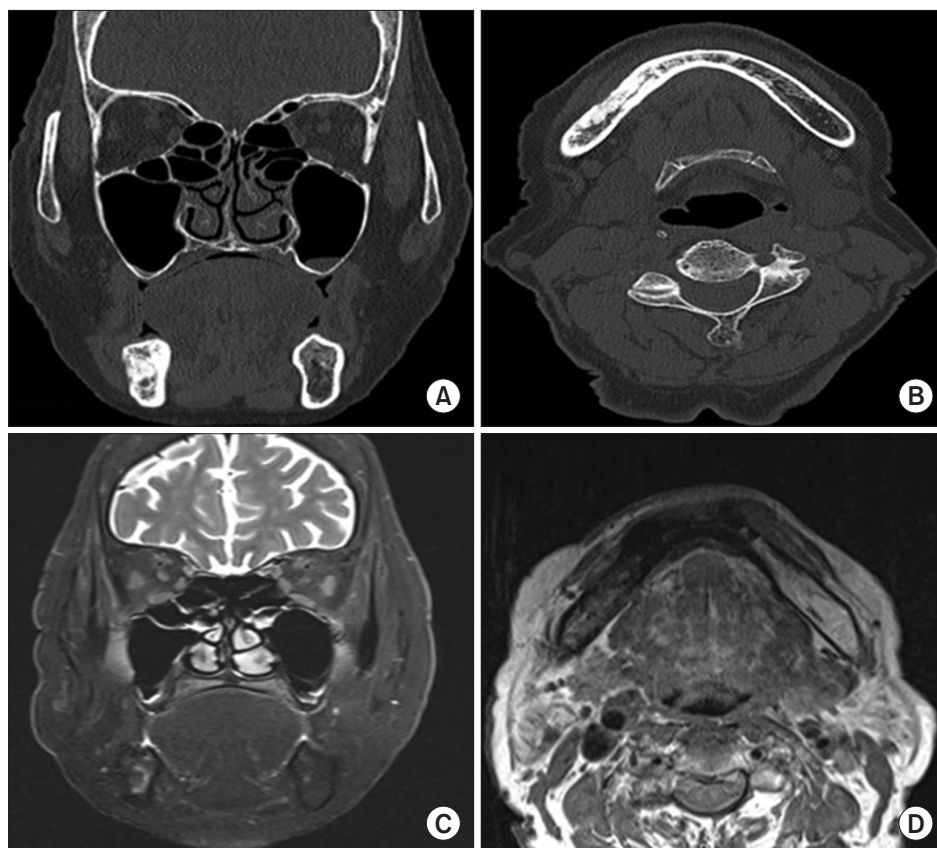


Fig. 6. Postoperative 9 months computed tomography (CT) and magnetic resonance imaging (MRI). The fistula tract was healed, there was no recurrence (A, C), and the bone at the operation site was regenerated (B, D) on CT and MRI images at postoperative 9 months.

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ant cells surrounding the necrotic bone with fibrosis was also noted.(Fig. 5)

Histologically, fluent foamy histiocytes, giant cells, and proliferative fibrosis provided the diagnosis of XGI.

There was no complaint of pain in the operation site at the follow-up appointment at 9 months. Since the IAN was involved in the tumor, a nerve sensation test was performed to assess sensory abnormalities in the right jaw area. Two-point discrimination and direction testing were performed at 2 weeks, 6 weeks, 4 months, and 7 months postoperatively; the results were within normal limits. Two-point discrimination was not reliable. The patient recognized two points at 15 mm on the bilateral cheek and malar area and at 13-17 mm

on both sides of the chin. She had 1-cm-sized region of hypoesthesia beneath the right side of the lower lip. Self-reported sensation was 50% at postoperative 2 weeks and improved to 80% at postoperative 7 months. The fistula tract was resolved after surgery, and bone regenerated well on CT and MRI images at postoperative 9 months.(Fig. 6)

III. Discussion

According to Krishna and Dayal⁵, only 3 cases of XGI of the head and neck region were published from January 2008 to April 2020, with one case each in the thyroid, thyroglossal cyst, and eye. There have been no cases involving the

mandibular bone area^{5,6}. XGI is a benign lesion that has been misdiagnosed as malignant disease as it is similar to infiltrative cancer on CT and MRI and presents as hot spots on 3-phase bone scan imaging. Histopathological examination is essential to diagnose XGI, characterized by foamy histiocytes and giant cells intermixed with other inflammatory cells and marked proliferative fibrosis. In this case, there were cortical irregularities on CT and hot spots on 3-phase bone scan imaging. XGI was diagnosed due to abundant foam cells on preoperative incisional biopsy.

Due to the rarity, XGI has no established treatment protocol. Most cases have been treated with curettage and bone grafting³. In some cases, XGI treatment involved only antibiotics. Therefore, antibiotic therapy with regular follow-up as a noninvasive treatment method should be considered before surgery^{3,7,8}. In our case, because the patient had complained of pain on the right mandible for almost 1 year, and the mass was approximately 4 cm in size, surgical treatment was performed.

Pre-antibiotic treatment was performed before surgery for 1 week. Surgical excision and neuroplasty were performed as the IAN was involved. However, there was no histopathological evidence that the mass affected the nerve system. In a previous case, the IAN canal was displaced inferiorly⁹. On the postoperative follow-up, there was no recurrence, and sensation of the chin had improved. However, nerve sensation testing remained unreliable because of the apparent age-related decline in the ability to discriminate two points¹⁰.

We present this case due to its rarity and curability. While XGI is benign, it can be misdiagnosed as a malignant bone lesion both clinically and radiographically. Histopathological examination of lesions is necessary to obtain an appropriate diagnosis. Further studies are needed on the recurrence rate and origin.

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Authors' Contributions

H.B. participated in data collection and wrote the manuscript. K.H.Y. participated in the study design and performed the case surgery. M.S.O. participated in the study design and coordination and helped to draft the manuscript. All authors

read and approved the final manuscript.

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Consent for Publishing Photographs

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

Conflict of Interest

No potential conflict of interest relevant to this article was reported.

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