

# Aesthetic Mandibular Angioplasty to Improve Patient Quality of Life in Chronic Recurrent Multifocal Osteomyelitis

Yun Yong Park, MD  
Bumjin Park, MD

**Summary:** Chronic recurrent multifocal osteomyelitis (CRMO) is a rare autoimmune disease that typically develops during adolescence and primarily affects women. CRMO primarily targets the bone in arms and legs, with sporadic occurrences in the mandible. CRMO is typically managed with medical treatment, and the efficacy of surgery remains controversial. Complications of surgery include massive bleeding and potential flare-up of CRMO symptoms. Herein, we report a patient with CRMO who had lesions in the bilateral rami of the mandible treated with aesthetic mandibular angioplasty. This is the first case of aesthetic mandibular angioplasty in a patient with CRMO who had bilateral rami involvement of the mandible. The patient began experiencing jaw pain accompanied by swelling and throbbing discomfort beneath the jawline at the age of 10. A pediatrician diagnosed CRMO, and the symptoms were controlled with nonsteroidal antiinflammatory drugs and immunosuppressants (infliximab, adalimumab). Aesthetic mandibular angioplasty was performed at our center because of mandibular hypertrophy. This procedure necessitated considerable removal of the spongy bone, raising concerns about potential massive intraoperative bleeding. Approximately 1.5 cm of the mandibular body was excised to reveal the cortical bone. Bleeding during surgery was not severe, rendering blood transfusions unnecessary. The patient was satisfied with the surgical results. This case indicates the feasibility of angioplasty for such cases. (*Plast Reconstr Surg Glob Open* 2024; 12:e5718; doi: [10.1097/GOX.0000000000005718](https://doi.org/10.1097/GOX.0000000000005718); Published online 8 April 2024.)

Chronic recurrent multifocal osteomyelitis (CRMO) is a rare autoimmune disease that typically develops during adolescence, and primarily affects women.<sup>1-3</sup> CRMO predominantly targets long bones, such as those in the arms and legs; however, rare occurrences of CRMO in the mandible have also been reported.<sup>1,2</sup> CRMO is characterized by aseptic inflammation within the bone, and is defined as an autoimmune-induced inflammatory disorder.<sup>2</sup> Management of CRMO generally involves medical treatment, emphasizing pain control as the primary therapeutic strategy.<sup>1-3</sup> Meanwhile, the efficacy of surgical

intervention for CRMO remains a topic of debate.<sup>4,5</sup> Bone resection, particularly when targeting a substantial portion of the spongy bone, can result in considerable bleeding during surgery. Furthermore, the possibility of symptom recurrence attributed to a CRMO flare-up after surgery is also a significant concern.<sup>6</sup> These complications explain why the prevailing surgical approach for CRMO involves limited resection of pathologically enlarged bones.<sup>5</sup>

Herein, we report the case of a patient with CRMO who had lesions in the bilateral rami of the mandible. The patient expressed considerable concern regarding the abnormal enlargement of the mandible; to alleviate this, we conducted a mandibular resection procedure aimed at enhancing cosmetic appearance. This is the first case of aesthetic mandibular angioplasty in a patient with CRMO who had bilateral rami involvement of the mandible. This study was approved by the institutional review board (approval no.: P01-202310-01-004).

## CASE REPORT

The patient began experiencing jaw pain, accompanied by swelling and throbbing discomfort beneath

From the Departments of Plastic and Reconstructive Surgery, iWELL Plastic Surgery Clinic, Seoul, Republic of Korea.

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**Fig. 1.** A, Preoperative photograph of the patient, showing the patient's large mandible. B, Postoperative photograph (6-month follow-up) displaying aesthetic improvement.

the jawline at the age of 10 years. Subsequently, in 2015, at age 11, the patient was diagnosed with mandibular osteomyelitis in the dental department of a general hospital in another country. Antibiotic therapy was initiated. Despite periods of improvement, recurrence occurred. However, in 2018, the patient's symptoms exacerbated; all management attempts were ineffective, even after pediatric intervention, and the patient was diagnosed with CRMO. The patient responded positively to nonsteroidal antiinflammatory drugs (NSAIDs) and immunosuppressants (infliximab, adalimumab), with reductions in symptoms.

The patient sought medical attention in August 2023 due to mandibular hypertrophy resulting from this disease (Fig. 1). Consequently, cosmetic mandibular angloplasty was performed at our hospital to improve this deformity.

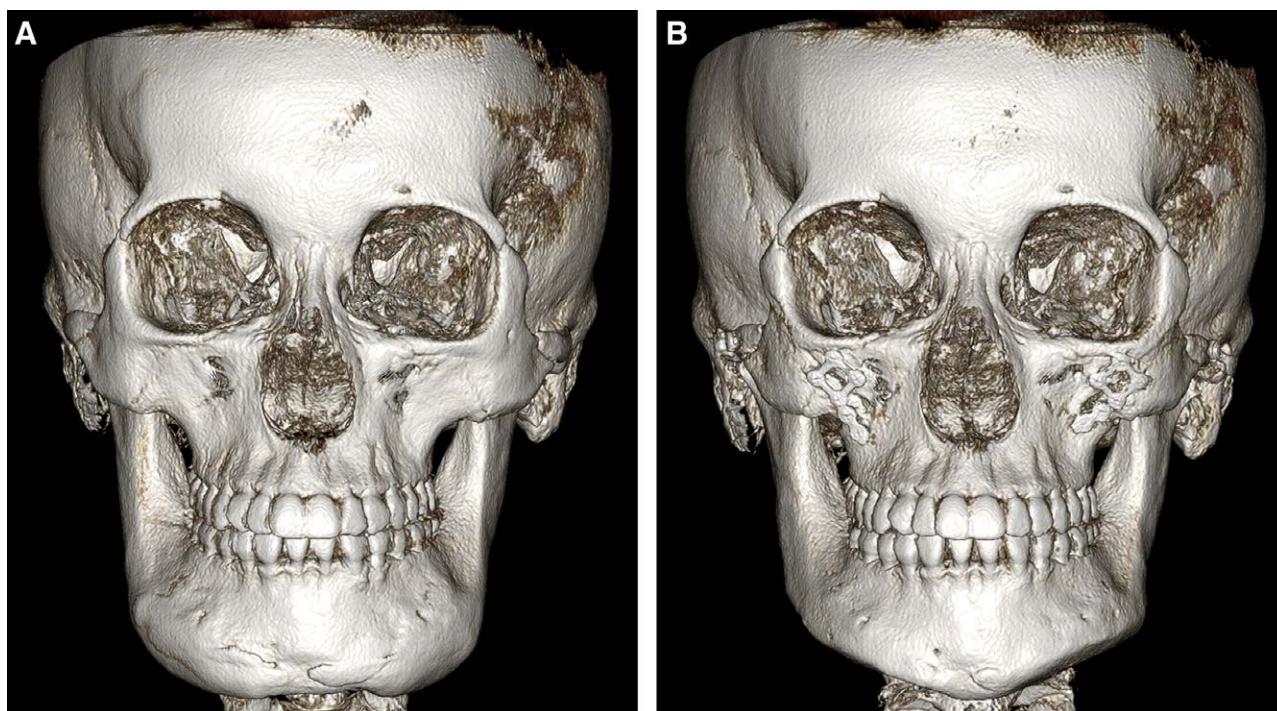
Abnormal thickening of the mandible was detected on computed tomography performed at our hospital (Fig. 2). In this case, a substantial portion of the spongy bone needed to be excised to perform mandibular aesthetic surgery. Consequently, owing to the anticipated potential for substantial bleeding during the operation, preemptive measures were taken to prepare for blood transfusion. The surgical procedure was performed under general anesthesia. We accessed the

mandible through an intraoral incision, taking care to avoid damage to the mental nerve. After exposing the lower border of the mandible, we designed the bone to be resected. The resected mandible measured approximately  $2 \times 8 \times 1.5$  cm on the right side and  $2 \times 5.2 \times 1.5$  cm on the left side.

On postoperative day 7, the patient did not have any complication, including pain. Preoperatively, her C-reactive protein (CRP) level was 0.2, and the erythrocyte sedimentation rate (ESR) was 10. In the immediate postoperative inflammation level assessment, CRP exhibited a slight elevation to 1.05, whereas the ESR increased to 31.

At the 6-month follow-up, substantial improvement in edema was noted, indicating aesthetic improvement (Fig. 1). There were no flare-ups of the autoimmune disease, and no notable complications occurred. Additionally, laboratory findings at the 6-month follow-up, including CRP(0.03), ESR(7), and white blood cell count were all within normal ranges.

Before surgery, the patient experienced psychological distress due to abnormal enlargement of the lower jaw. We measured quality of life with Rosenberg self-esteem scale. The patient's Rosenberg self-esteem scale score increased from 18 before surgery, indicating mild self-esteem decline, to 22 after surgery, demonstrating improvement.



**Fig. 2.** A, Preoperative computed tomography, showing the abnormal thickening of mandible. B, Postoperative computed tomography, showing that a substantial amount of the mandible was resected.

### DISCUSSION

Most patients with CRMO are cured with medical treatments without significant sequelae; however, in some patients, sequelae may persist.<sup>7,8</sup> Indeed, 20% of patients in the study by Catalano-Pons et al<sup>7</sup> and 7% in the review by Schultz et al<sup>8</sup> experienced long-term sequelae. Surgical intervention is required in cases where symptoms persist; however, surgical management of CRMO remains controversial.<sup>4,5</sup> Concerns mainly center on potential CRMO symptom recurrence, along with concerns about bleeding complications. In general, it is known that autoimmune disease flare-ups are associated with T-cell activation,<sup>9</sup> with one study showing that flare-ups are highly likely to occur because T cells are activated after stressful situations, including surgery.<sup>6</sup> This has led to most surgeons preferring limited bone removal as the optimal surgical option. However, bone hypertrophy that occurs as a result of disease progression can significantly affect a patient's quality of life. In particular, when hypertrophy affects facial areas such as the mandible, it can lead to severe psychological distress. Therefore, adopting an aesthetic surgical approach is essential for patients with CRMO.

After performing surgery on the CRMO patient, we took several factors into consideration. Firstly, we assessed whether there was any pain due to CRMO recurrence. Additionally, we conducted laboratory work before surgery, at 7 days postoperative, and at 6 months postoperative to check for signs of inflammation. White blood cell count, CRP, and ESR were among the inflammatory markers we monitored. Furthermore, considering that

NSAIDs are the treatment of choice for managing symptoms in CRMO patients, we used NSAIDs for pain relief postoperatively.

This case highlights the relative safety and dependability of surgical treatment in patients with CRMO and bone hypertrophy. Despite concerns regarding massive bleeding and CRMO symptom resurgence, bleeding was controlled, and the patient experienced no notable CRMO symptom recurrences. This case further underscores the necessity for aesthetic mandibular resection in patients with sequelae of hypertrophy.

### CONCLUSIONS

Historically, there have been no reports of CRMO with bilateral rami involvement, and attempts to actively address aesthetic concerns have been scarce worldwide. In this case, we successfully performed aesthetic mandibular angioplasty in a patient with bilateral rami involvement for the first time.

*Bumjin Park, MD*

Departments of Plastic and Reconstructive Surgery  
iWELL Plastic Surgery Clinic  
4th EGI Building, 843 Nonhyunro  
Gangnamgu, Seoul 06031, Republic of Korea  
E-mail: [bumjinparkmd@gmail.com](mailto:bumjinparkmd@gmail.com)

### DISCLOSURE

*The authors have no financial interest to declare in relation to the content of this article.*

**PATIENT CONSENT**

*The patient provided written consent for the use of her images.*

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