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# Chronic Mandibular Actinomycosis with Osteomyelitis: A 10-Year Follow-up Case Report of Multidisciplinary Management

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#### **Abstract**

**Background:** Cervicofacial actinomycosis is a chronic granulomatous infection caused by Actinomyces species that can lead to significant morbidity if not adequately treated. Mandibular osteomyelitis represents an uncommon but serious complication that requires aggressive multidisciplinary management. **Case Presentation:** We present a 39-year-old male with chronic mandibular actinomycosis complicated by osteomyelitis, presenting with severe trismus, facial asymmetry, and multiple draining sinuses. The patient underwent buccal decortication combined with prolonged antimicrobial therapy, resulting in successful clinical resolution.

**Conclusion:** This case highlights the challenging nature of chronic actinomycosis management and demonstrates the effectiveness of combined surgical debridement and targeted antimicrobial therapy in achieving favorable outcomes.

**Keywords:** Actinomycosis, mandibular osteomyelitis, buccal decortication, cervicofacial infection, chronic granulomatous infection

#### Introduction

Actinomyces species are gram-positive, anaerobic bacteria that naturally inhabit the oral cavity, oropharynx, and digestive tract as normal flora<sup>1</sup>. When pathogenic conditions arise, these organisms typically cause infections characterized by gradual onset, prolonged duration, and a tendency to form chronic granulomatous lesions with characteristic sulfur granules<sup>2,3</sup>. Cervicofacial actinomycosis, most frequently caused by *Actinomyces israelii*, accounts for approximately 50-60% of all actinomycotic infections and predominantly affects soft tissue structures, with bone involvement being relatively uncommon <sup>4,5</sup>.

The development of actinomycotic disease typically requires disruption of mucosal barriers through antecedent infection, trauma, or surgical intervention, allowing these normally commensal bacteria to penetrate deeper anatomical structures and establish progressive infection<sup>6</sup>. The characteristic pattern of disease extension involves continuous local tissue invasion with formation of multiple draining sinuses rather than distant hematogenous spread<sup>7</sup>.



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Mandibular osteomyelitis caused by Actinomyces represents a rare complication that poses significant diagnostic and therapeutic challenges<sup>8</sup>. The condition often mimics other chronic infectious processes, malignancies, or inflammatory conditions, leading to delayed diagnosis and treatment<sup>9,10</sup>. Establishing an accurate diagnosis proves difficult due to clinical and radiological similarities with various other conditions, including chronic suppurative osteomyelitis, malignancy, and other granulomatous infections<sup>11</sup>.

The management of chronic actinomycotic osteomyelitis requires a multidisciplinary approach combining surgical debridement with prolonged antimicrobial therapy<sup>12,13</sup>. This case report presents our clinical experience managing this rare condition and describes the therapeutic approach that led to successful outcomes in a patient with a 10-year history of chronic mandibular actinomycosis.

## Case Report

#### **Patient Information and Clinical Presentation**

A 39-year-old male presented to the Department of Oral and Maxillofacial Surgery with a chief complaint of severe pain on the left side of the lower jaw associated with markedly reduced mouth opening. The patient reported chronic symptoms spanning over 5 years with recent acute exacerbation significantly affecting his ability to eat and perform activities of daily living.

## **Medical History**

The patient had no known drug allergies and no significant systemic medical conditions. However, he had an extensive surgical history related to his mandibular actinomycosis:

**2015:** Initial presentation and diagnosis

- Underwent exploration and excision biopsy of mandibular lesion
- Histopathological examination confirmed actinomycosis

**2016:** Disease progression requiring surgical intervention

- Developed recurrent pain and swelling in the left mandibular region
- Underwent buccal decortication under general anesthesia

2016-2021: Long-term medical management

- Continuous medical management for recurrent episodes
- Treatment included prolonged antibiotic therapy and bisphosphonate therapy for associated osteomyelitis

#### **Physical Examination**

On admission, vital signs were stable (temperature 98.5°F, pulse 68 bpm, respiratory rate 20/min, blood pressure 125/80 mmHg). Systemic examination revealed normal cardiovascular and respiratory systems. **Extraoral examination** revealed gross facial asymmetry with diffuse swelling on the left side of the face. Multiple healed scar wounds with active drainage sites were observed along the left lower border of the mandible and chin region. Palpation revealed tenderness over the scarred areas. Mandibular deviation toward the right side was noted during attempted mouth opening.



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**Intraoral examination** showed severe trismus with mouth opening restricted to approximately 5mm. Dental occlusion remained stable despite the limited opening, and no step deformities were observed. Vestibular examination showed no tenderness or obliteration.



**Laboratory Investigations** 

Comprehensive laboratory studies performed on admission (November 5, 2024) revealed:

**Hematological parameters:** Hemoglobin 14.2 g/dL, total white blood cell count  $8.88 \times 10^3/\mu L$  with normal differential, platelet count  $330 \times 10^3/\mu L$ 

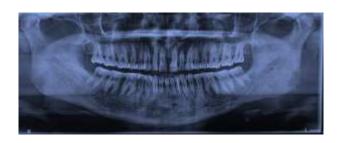
**Biochemical analysis:** Normal renal function (blood urea 14 mg/dL, serum creatinine 0.7 mg/dL), normal liver function tests, slightly elevated serum sodium (149 mmol/L), and normal random blood glucose (119 mg/dL)

Coagulation studies: Normal prothrombin time, activated partial thromboplastin time, and international normalized ratio

**Infectious disease screening:** Negative serology for HIV, Hepatitis B, and Hepatitis C; negative Gene-Xpert testing for Mycobacterium tuberculosis

## **Imaging Studies**

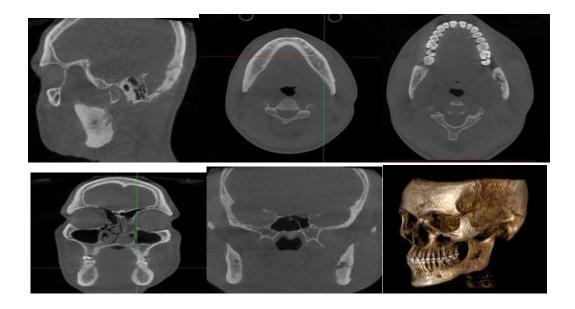
**Orthopantomogram** showed irregularities in the left mandibular angle region consistent with chronic osteomyelitis.





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CT and MRI scans of the head and neck region revealed heterogeneous enhancement in the muscles of the left infratemporal/masticator space with a thin plaque-like hyperintense collection in the left parapharyngeal space, suggestive of infective/inflammatory etiology.



Chest X-ray and high-resolution CT of chest: No significant abnormalities detected Echocardiogram: Normal cardiac chambers with good left ventricular systolic function (ejection fraction 60%)

## **Diagnosis**

Based on the clinical presentation, physical examination findings, imaging studies, and documented histopathological evidence, a diagnosis of chronic actinomycotic osteomyelitis of the left mandible was established.

# **Treatment and Management**

## **Surgical Intervention**

The patient underwent buccal decortication of the left mandibular angle region under general anesthesia on November 15, 2024. The procedure was performed using a submandibular approach with the following key steps:

- 1. Standard aseptic preparation and draping
- 2. Nasal intubation and local anesthetic infiltration
- 3. Layer-by-layer dissection through submandibular approach
- 4. Identification and removal of fibrous tissue and granulation tissue
- 5. Ligation of facial artery for hemostasis
- 6. Release of pterygomassetic sling to expose mandibular angle
- 7. Creation of bur holes for drainage
- 8. Thorough buccal decortication to remove infected bone
- 9. Copious irrigation with metronidazole and 3% saline solution
- 10. Local application of tetracycline
- 11. Placement of surgical drain



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## 12. Layer-wise closure with non-absorbable sutures



The procedure was completed without complications.

# **Medical Management**

## Antimicrobial therapy:

- Intravenous gentamicin 80mg twice daily
- Intravenous clindamycin 600mg twice daily
- Oral cotrimoxazole (Bactigram DS) twice daily for extended duration

#### **Supportive care:**

- Intravenous pantoprazole 40mg once daily
- Oral pentoxifylline 400mg twice daily
- Topical neomycin ointment
- Chlorhexidine mouthwash four times daily

## **Clinical Course and Outcomes**

The patient's postoperative course was uneventful. Regular follow-up monitoring at 1 week, 3 weeks, 1 month, 3 months, and 6 months demonstrated significant clinical improvement with controlled pain levels, stable wound healing, and improved functional status. Laboratory investigations showed normalization of serum electrolytes and maintenance of stable renal function parameters.





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#### Discussion

This case illustrates the complex management challenges associated with chronic actinomycotic osteomyelitis of the mandible. Several important clinical and therapeutic considerations emerge from this experience.

## **Diagnostic Challenges**

The diagnosis of mandibular actinomycosis remains challenging due to its clinical and radiological similarity to other chronic infectious conditions, malignancies, and inflammatory processes<sup>14,15</sup>. The condition often presents with nonspecific symptoms including pain, swelling, trismus, and draining sinuses, which can be mistaken for other pathological processes<sup>16</sup>. In this case, the patient's documented histopathological evidence and characteristic clinical presentation facilitated diagnosis, emphasizing the importance of tissue biopsy in establishing definitive diagnosis<sup>17</sup>.

#### **Surgical Management Principles**

Buccal decortication represents a well-established surgical approach for managing mandibular osteomyelitis, particularly in cases involving the mandibular angle and ramus<sup>18,19</sup>. The procedure aims to remove infected and necrotic bone tissue, improve local vascularization, eliminate bacterial reservoirs, and facilitate antibiotic penetration into the affected tissue<sup>20</sup>. The surgical technique employed in this case, including thorough debridement, copious irrigation, local antibiotic application, and adequate drainage, follows established principles for managing chronic osteomyelitis<sup>21,22</sup>.

The decision to perform buccal decortication was based on the extent of bone involvement, failure of previous conservative management, and the need for adequate surgical access to remove infected tissue<sup>23</sup>. The submandibular approach provided excellent visualization and access to the affected mandibular angle region while minimizing the risk of injury to vital structures<sup>24</sup>.

#### **Antimicrobial Therapy Considerations**

The antimicrobial regimen utilized in this case reflects current understanding of actinomycosis treatment principles<sup>25,26</sup>. Clindamycin demonstrates excellent activity against anaerobic bacteria, including Actinomyces species, and achieves good bone penetration<sup>27</sup>. Gentamicin provides broad-spectrum coverage against potential secondary bacterial infections commonly associated with chronic osteomyelitis<sup>28</sup>. The inclusion of cotrimoxazole for extended oral therapy is consistent with recommendations for long-term suppressive therapy in chronic actinomycosis<sup>29,30</sup>.



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The duration of antimicrobial therapy in actinomycosis typically requires prolonged treatment courses, often extending for several months to years, depending on the extent of disease and response to therapy<sup>31</sup>. This extended treatment duration is necessary due to the organism's slow growth characteristics and the tendency for infection to recur if treatment is inadequate<sup>32</sup>.

### **Multidisciplinary Approach**

The successful management of this case required coordination between multiple medical specialties, including oral and maxillofacial surgery, internal medicine, and infectious disease consultation<sup>33</sup>. This multidisciplinary approach ensures comprehensive evaluation and management of both local and systemic aspects of the condition<sup>34</sup>.

## **Clinical Outcomes and Prognosis**

The favorable outcome achieved in this case demonstrates the effectiveness of combined surgical and medical management when appropriately applied<sup>35</sup>. The absence of postoperative complications and stable follow-up parameters indicate successful treatment response and emphasize the importance of adequate surgical debridement combined with targeted antimicrobial therapy<sup>36</sup>.

Long-term follow-up remains crucial in patients with chronic actinomycosis due to the potential for disease recurrence and the need for ongoing monitoring of treatment response<sup>37,38</sup>.

#### Conclusion

Chronic actinomycotic osteomyelitis of the mandible represents a challenging clinical condition that requires aggressive multidisciplinary management. This case demonstrates that successful outcomes can be achieved through prompt recognition and diagnosis, appropriate surgical intervention with thorough debridement, targeted antimicrobial therapy with adequate duration, comprehensive postoperative care, and patient compliance with treatment protocols.

The favorable outcome supports the continued use of buccal decortication combined with prolonged antimicrobial therapy as an effective treatment strategy for mandibular actinomycotic osteomyelitis. However, further research is needed to optimize treatment protocols and identify factors associated with successful long-term outcomes in patients with chronic cervicofacial actinomycosis.

#### **Patient Consent**

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

#### References

- 1. Smego RA Jr, Foglia G. Actinomycosis. Clin Microbiol Rev. 1998;11(3):439-69.
- 2. Pulverer G, Schütt-Gerowitt H, Schaal KP. Human cervicofacial actinomycoses: microbiological data for 1997 cases. Clin Microbiol Rev. 2003;16(1):37-58.
- 3. Valour F, Sénéchal A, Dupieux C, Karsenty J, Lustig S, Breton P, et al. Actinomycosis: etiology, clinical features, diagnosis, treatment, and management. Infect Drug Resist. 2014;7:183-97.
- 4. Brook I. Actinomycosis: diagnosis and management. South Med J. 2008;101(10):1019-23.



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- 5. Russo TA. Agents of actinomycosis. In: Mandell GL, Bennett JE, Dolin R, editors. Mandell, Douglas, and Bennett's Principles and Practice of Infectious Diseases. 7th ed. Philadelphia: Churchill Livingstone Elsevier; 2010. p. 3209-19.
- 6. Könönen E, Wade WG. Actinomyces and related organisms in human infections. Clin Microbiol Rev. 2015;28(2):419-42.
- 7. Metgud S, Sumati, Astekar M. Cervicofacial actinomycosis: a clinicopathologic study. Ann Maxillofac Surg. 2012;2(2):121-6.
- 8. Bartkowski SB, Zapała J, Heczko P. Actinomycotic osteomyelitis of the mandible: review of 15 cases. J Craniomaxillofac Surg. 1998;26(3):155-60.
- 9. Ha YS, Choi WI, Cheon YI, Kwon YS. Pulmonary actinomycosis with endobronchial involvement: a rare cause of endobronchial polypoid lesions. Intern Med. 2010;49(18):1977-82.
- 10. Moturi K, Prasad S, Devaraj U, Stroup JS. Cervicofacial actinomycosis: an elusive diagnosis. Case Rep Dent. 2013;2013:740172.
- 11. Wong VK, Turmezei TD, Weston VC. Actinomycosis. BMJ. 2011;343:d6099.
- 12. Boyanova L, Kolarov R, Mateva L, Markovska R, Mitov I. Actinomycosis: a frequently forgotten disease. Future Microbiol. 2015;10(4):613-28.
- 13. Acevedo F, Baudrand R, Letelier LM, Gaete P. Actinomycosis: a great pretender. Case reports of unusual presentations and a review of the literature. Int J Infect Dis. 2008;12(4):358-62.
- 14. Miller M, Haddad AJ. Cervicofacial actinomycosis. Oral Surg Oral Med Oral Pathol Oral Radiol Endod. 1998;85(5):496-508.
- 15. Sharkawy AA. Cervicofacial actinomycosis and mandibular osteomyelitis. Infect Dis Clin North Am. 2007;21(2):543-56.
- 16. Ficarra G, Beninati F. Unusual clinical presentations of oral actinomycosis. J Oral Maxillofac Surg. 2002;60(10):1211-4.
- 17. Schaal KP, Lee HJ. Actinomycete infections in humans--a review. Gene. 1992;115(1-2):201-11.
- 18. Kumar VV, Jacob PC, Ebenezer S, Kuriakose AM, Keerthi G. Buccal cortical plate perforation as a marker of bisphosphonate-related osteonecrosis: a preliminary radiographic study. Oral Surg Oral Med Oral Pathol Oral Radiol. 2013;115(6):749-54.
- 19. Marx RE. Osteoradionecrosis: a new concept of its pathophysiology. J Oral Maxillofac Surg. 1983;41(5):283-8.
- 20. Lew DP, Waldvogel FA. Osteomyelitis. Lancet. 2004;364(9431):369-79.
- 21. Cierny G 3rd, Mader JT, Penninck JJ. A clinical staging system for adult osteomyelitis. Clin Orthop Relat Res. 2003;(414):7-24.
- 22. Lima AL, Oliveira PR, Carvalho VC, Saconi ES, Cabrita HB, Rodrigues MB. Recommendations for the treatment of osteomyelitis. Braz J Infect Dis. 2014;18(5):526-34.
- 23. Brady BA, Leid JG, Camper AK, Costerton JW, Shirtliff ME. Identification of Staphylococcus aureus proteins recognized by the antibody-mediated immune response to a biofilm infection. Infect Immun. 2006;74(6):3415-26.
- 24. Ellis E 3rd, Hupp JR, Tucker MR. Contemporary Oral and Maxillofacial Surgery. 6th ed. St. Louis: Elsevier Mosby; 2014.
- 25. Bennett JE, Dolin R, Blaser MJ. Mandell, Douglas, and Bennett's Principles and Practice of Infectious Diseases. 8th ed. Philadelphia: Elsevier Saunders; 2015.
- 26. Yates AJ. Postoperative medical management. Orthop Clin North Am. 2010;41(1):25-33.



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- 27. Baddour LM. Long-term suppressive antimicrobial therapy for intravascular device-related infections. Am J Med Sci. 2001;322(4):209-12.
- 28. Gilbert DN, Chambers HF, Eliopoulos GM, Saag MS, Pavia AT. The Sanford Guide to Antimicrobial Therapy 2019. 49th ed. Sperryville: Antimicrobial Therapy Inc; 2019.
- 29. Sudhakar SS, Ross JJ. Short-term treatment of actinomycosis: two cases and a review. Clin Infect Dis. 2004;38(3):444-7.
- 30. Bennhoff DF. Actinomycosis: diagnostic and therapeutic considerations and a review of 32 cases. Laryngoscope. 1984;94(9):1198-217.
- 31. Weese WC, Smith IM. A study of 57 cases of actinomycosis over a 36-year period. A diagnostic 'failure' with good prognosis after treatment. Arch Intern Med. 1975;135(12):1562-8.
- 32. Brown JR. Human actinomycosis. A study of 181 subjects. Hum Pathol. 1973;4(3):319-30.
- 33. Mabeza GF, Macfarlane J. Pulmonary actinomycosis. Eur Respir J. 2003;21(3):545-51.
- 34. Garner JP, Macdonald M, Kumar PK. Abdominal actinomycosis. Int J Clin Pract. 2007;61(4):603-8.
- 35. Kinnear WJ, MacFarlane JT. A survey of thoracic actinomycosis. Respir Med. 1990;84(1):57-9.
- 36. Hsieh MJ, Liu HP, Chang JP, Chang CH. Thoracic actinomycosis. Chest. 1993;104(2):366-70.
- 37. Fiorino AS. Intrauterine contraceptive device-associated actinomycotic abscess and actinomycosis: a clinicopathologic study of 69 cases with review of the literature. Hum Pathol. 1996;27(4):334-43.
- 38. Cope VZ. Actinomycosis of salivary glands. Br J Surg. 1938;26(102):255-70.