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Case Report

Pediatric cervicofacial actinomycosis presenting with mandibular osteomyelitis: a diagnostic and therapeutic challenge and literature review

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ABSTRACT

Background. Actinomycotic osteomyelitis is a rare, chronic infection caused by *Actinomyces* species, anaerobic bacteria normally found in the oral flora. Mandibular involvement is uncommon and may mimic malignancies, complicating diagnosis.

Case Presentation. A 15-year-old previously healthy male presented with painful neck swelling and trismus. Examination revealed a firm mass at the angle of the mandible. Imaging suggested osteomyelitis but raised concern for malignancy. Surgical drainage was performed, and histopathology confirmed actinomycotic infection. The patient initially received empirical intravenous vancomycin, cefotaxime, and metronidazole. Due to persistent fever and elevated inflammatory markers, the regimen was switched to teicoplanin plus piperacillin/tazobactam. Therapy was subsequently escalated to meropenem due to recurrent fever and based on magnetic resonance imaging findings suggestive of osteomyelitis, then vancomycin was replaced with teicoplanin because of vancomycin-associated nephrotoxicity. Following clinical improvement, the patient was discharged on oral amoxicillin/clavulanate to complete a total of six weeks of therapy.

Discussion. Pediatric mandibular actinomycotic osteomyelitis is extremely rare. Its indolent course often mimics tumors or granulomatous disease. In this case, delayed diagnosis and nonspecific imaging findings led to initial misinterpretation. Surgical intervention played a key role in diagnosis and treatment. Early recognition and combined medical-surgical management are crucial to avoid complications. This case highlights the importance of considering infectious causes in mandibular masses and underscores the diagnostic challenges associated with them.

Key words: actinomycosis, *Actinomyces*, mandibular osteomyelitis, pediatric.

Actinomycosis is a chronic infection caused by anaerobic, Gram-positive filamentous bacteria of the genus *Actinomyces*. *Actinomyces* are normally found in the oropharynx, gastrointestinal tract, and genitourinary tract. Under certain conditions, these bacteria can become pathogenic.[1,2] Pediatric actinomycosis is rare and often presents with nonspecific symptoms that mimic other infections or tumors, complicating diagnosis. Actinomycotic infections typically involve primary soft tissues in chronic inflammatory conditions, rarely

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affecting the bones.[3] Bacterial cultures and histopathological examination are fundamental to establishing an accurate diagnosis.[4]

This case report presents a rare pediatric cervicofacial actinomycosis involving the mandible, highlighting the importance of imaging, biopsy, and multidisciplinary treatment for successful outcomes.

Case Presentation

A 15-year-old male patient with no known comorbidities presented to the clinic with progressively enlarging, painful neck swelling, difficulty opening his mouth and restricted neck movement over the past 10 days. On physical examination, hypertrophic tonsils and dental caries in the right mandibular region were noted. A firm-to-hard, diffuse erythematous swelling was observed in the paratracheal region, with restricted mobility in all directions.

Laboratory tests revealed an elevated C-reactive protein (CRP) level of 99 mg/L, erythrocyte sedimentation rate (ESR) of 38 mm/h, white blood cell (WBC) count of 8.13 $\times 10^3/\mu L$, hemoglobin level of 12.7 g/dL, absolute neutrophil count (ANC) of 5.18 $\times 10^3/\mu L$, and platelet count of 210000/μL

Ultrasound (US) examination showed thickened, inflamed subcutaneous tissue in the midline of the neck, with a 19x11 mm hypoechoic collection suggestive of phlegmon or abscess. A computed tomography (CT) scan revealed increased density and inflammatory changes extending from the anterior neck to the left mandibular region, consistent with deep neck infection. Mild density changes in the bone marrow of the left mandibular ramus raised concerns for osteomyelitis.

Empirical intravenous therapy with vancomycin, cefotaxime, and metronidazole was initiated (Fig. 1). An otolaryngology consultation resulted in the percutaneous drainage of approximately 3 mL of purulent material. On the third day of hospitalization, as the swelling and restricted mobility persisted, interventional radiology performed ultrasound-guided percutaneous drainage of the abscess.

On the fourth day, due to an inadequate clinical response, cefotaxime was discontinued and replaced with piperacillin/tazobactam, while vancomycin therapy was continued. On the seventh day, the patient developed vomiting following vancomycin administration, and an elevation in serum creatinine to 0.94 mg/dL raised concerns for vancomycin-associated nephrotoxicity. Vancomycin was replaced with teicoplanin.

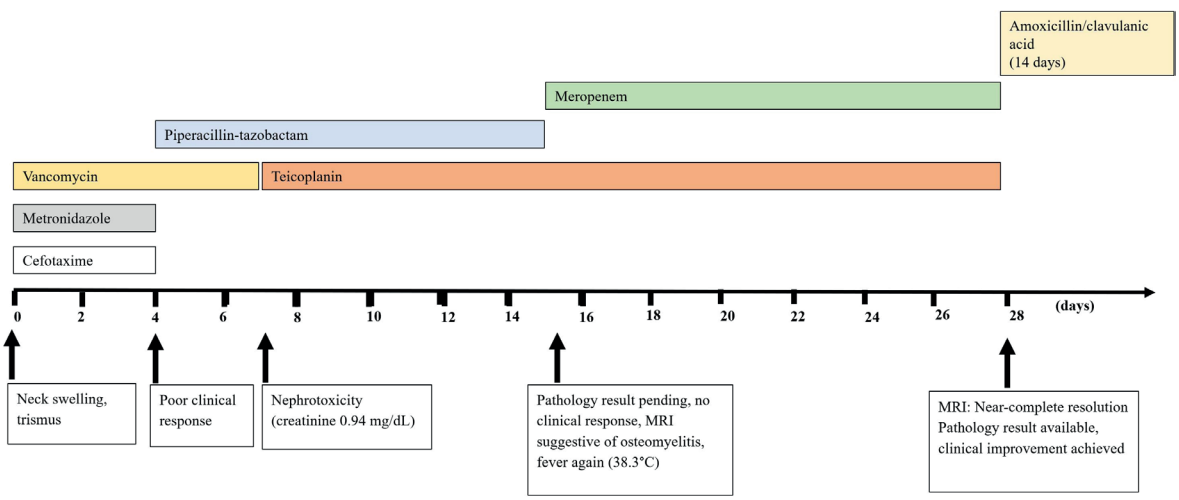

Fig. 1. Chronological summary of the clinical course and treatment.

Magnetic resonance imaging (MRI) revealed extensive anterior cervical soft tissue inflammation extending from the submental region to the thyroid level. A deep-seated lesion measuring 13×9 mm adjacent to the hyoid bone was identified, consistent with a phlegmon. Multiple enlarged cervical lymph nodes were observed. Additionally, signal alterations around the roots of the fourth and fifth mandibular molars raised suspicion for mandibular osteomyelitis (Fig. 2).

On the seventh day, cervical mobility had returned to normal. US demonstrated residual cellulitis and a 10×4 mm hypoechoic area consistent with a resolving phlegmon. On day 15, the patient developed a fever of 38.3 °C, although no new infectious focus was identified. Laboratory results were as follows: creatinine 0.76 mg/dL, CRP 7.5 mg/L, ESR 38 mm/h, procalcitonin 0.21 ng/mL, WBC $13\times10^3/\mu L$. Then, inflammatory markers worsened, with CRP increasing to 47 mg/L and WBC count rising to

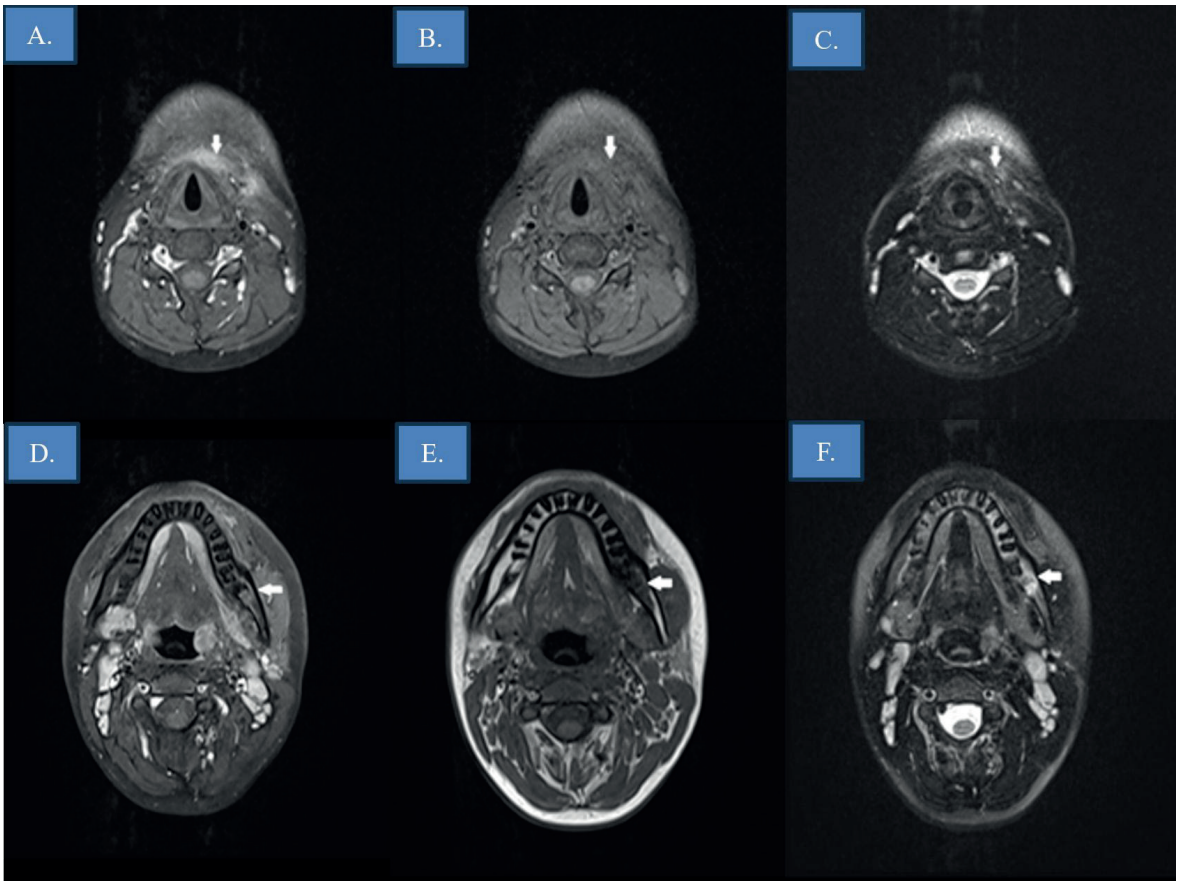

Fig. 2. Magnetic resonance imaging (MRI) showing altered signal intensity in the left mandibular ramus consistent with osteomyelitis and a 13 × 9 mm phlegmon in the anterior neck adjacent to the hyoid bone **A and B.** Axial T1-weighted fat-suppressed post-contrast images demonstrates diffuse inflammatory soft-tissue changes, consistent with phlegmon (arrows). **C.** Axial T2-weighted fat-suppressed MRI demonstrates a diffuse area of high signal intensity in the subcutaneous and deep cervical soft tissues (arrow), consistent with a phlegmon. **D.** Axial T1-weighted fat-suppressed post-contrast image demonstrates intense enhancement and marrow signal abnormality along the mandibular body (arrow), consistent with osteomyelitis. **E.** Axial T1-weighted MRI shows low signal intensity in the cortical and medullary portions of the mandibular bone (arrow), consistent with osteomyelitis. Adjacent soft-tissue inflammatory changes are also present. **F.** Axial T2-weighted fat-suppressed image shows hyperintense marrow signal and surrounding soft-tissue edema along the mandibular body (arrow), consistent with osteomyelitis.

15×10³/μL. Due to ongoing intermittent fever and the presence of suspected osteomyelitis, piperacillin/tazobactam was discontinued, and meropenem therapy was initiated.

Wound cultures showed no bacterial growth. Direct Gram staining of the drained fluid did not demonstrate identifiable organisms. Histopathological examination of the abscess material revealed abundant polymorphonuclear leukocytes and histiocytes, along with nonspecific inflammatory infiltrates and vascular proliferation. A cluster of filamentous microorganisms morphologically suggestive of *Actinomyces* species was identified, further supported by positive periodic acid–Schiff (PAS) staining (Fig. 3). Modified acid-fast staining was negative. This finding argues against *Nocardia* spp., which typically demonstrate partial acid-fast positivity due to the presence of mycolic acids in their cell walls. Therefore, the absence of acid-fast staining favored *Actinomyces* over *Nocardia* in the differential diagnosis. Blood cultures remained negative. Upon consultation with the microbiology department, it was noted that anaerobic cultures had not been performed. On day 21, follow-up ultrasonography demonstrated marked regression of the inflammatory process, with only a 9×3 mm residual hypoechoic lesion in the anterior cervical region. In light of the confirmed *Actinomyces* infection and mandibular osteomyelitis, the patient completed a total of four weeks of intravenous antibiotic therapy. Although *Actinomyces* species are highly susceptible to penicillin, de-escalation was not pursued in our patient due to the severity of infection, suspected mandibular osteomyelitis, and ongoing clinical instability at the time of histopathologic confirmation. Broad-spectrum therapy was therefore maintained to ensure adequate coverage until clinical resolution was achieved.

Follow-up MRI on day 28 demonstrated near-complete resolution of cellulitis, with no evidence of abscess formation. The patient was discharged on oral antibiotic therapy to

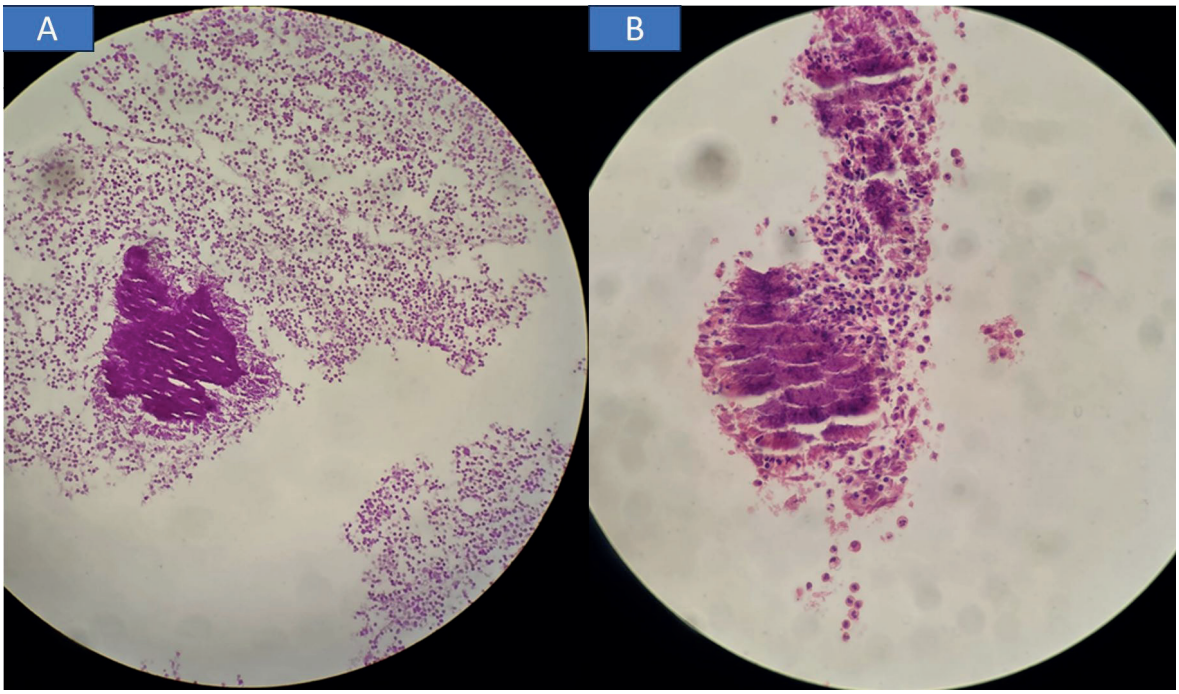

Fig. 3. Histopathological findings of the abscess material. **A.** Compact basophilic colony of filamentous microorganisms with radiating architecture, morphologically suggestive of *Actinomyces* species (H&E, original magnification x40) **B.** Filamentous bacterial colony highlighted by positive periodic acid–Schiff (PAS) staining.

complete a total of six weeks of treatment (Fig. 1). A written informed consent was obtained from the parents of the patient for this publication.

Discussion

Pediatric actinomycosis is a rare but challenging infection, often presenting as deep neck infections caused by *Actinomyces* species, Gram-positive, filamentous, anaerobic bacteria found in the oral cavity, gastrointestinal, and genitourinary tracts.[4] While these infections typically involve soft tissues with a chronic course, bone involvement is uncommon.[5] Poor oral hygiene and antecedent oropharyngeal disease increase susceptibility; in our patient, the presence of untreated dental caries likely contributed to the disruption of mucosal barriers, facilitating the invasion of *Actinomyces* and subsequent mandibular involvement.[6] A 16-year retrospective study by Chew et al. identified 14 pediatric cases, mostly cervicofacial, followed by thoracic and abdominopelvic forms.[6] The mandible's poor vascularity may contribute to its susceptibility.[7,8] A broader review of the literature reveals that pediatric cases of actinomycotic osteomyelitis are extremely rare, with only a limited number published to date.[9-20] These cases, summarized in Table I, highlight the variability in age, infection site, microbiologic findings, and management strategies and emphasize the diagnostic challenges associated with this unusual clinical entity.

This case is notable for its acute presentation, with trismus and a rapidly enlarging mass—features that mimicked malignancy. This aligns with prior studies showing that pediatric actinomycosis may resemble tumors or granulomatous diseases, leading to misdiagnosis or delayed treatment.[6] Although *Actinomyces* infections are typically chronic, they may present acutely in children, complicating diagnosis. Clinical and radiological findings are often nonspecific and may mimic malignancy or chronic infections. Chew et al.[6] similarly reported that pediatric actinomycosis can resemble neoplastic or granulomatous diseases, leading to delayed diagnosis and treatment.

Despite negative wound cultures, the diagnosis was ultimately confirmed via histopathologic analysis. This reflects existing challenges in isolating *Actinomyces* due to their slow growth and anaerobic nature.[4,7,8] Consequently, histopathology—particularly the detection of sulfur granules and filamentous Gram-positive organisms—remains a cornerstone in diagnosis. Although histopathology confirmed the presence of *Actinomyces,* anaerobic cultures were not performed in this case. This major limitation resulted in delays in both definitive diagnosis and initiation of targeted therapy, underscoring the critical role of anaerobic cultures when actinomycosis is suspected.

Surgical management of actinomycotic osteomyelitis, including curettage, sequestrectomy, and peripheral osteotomy, plays a critical role in both diagnosis and treatment.[21] Although penicillin is generally effective against *Actinomyces*, broad-spectrum antibiotics were initially preferred in our patient due to the infection's severity and poor early response. Chew et al.[6] recommend tailoring antibiotic regimens to the infection's extent, location, and course, supporting this approach.

Actinomyces species are slow-growing, with low metabolic activity, and form dense tissue abscesses and sinus tracts, which limit antibiotic penetration and delay clinical response.[2] Consequently, prolonged antibiotic therapy is required to achieve adequate tissue concentrations and prevent relapse. The standard regimen consists of high-dose intravenous penicillin G for 2–6 weeks.[1,4] This extended treatment is particularly important in cases with deep tissue or bone involvement, such as mandibular osteomyelitis. Shorter courses have been associated with higher relapse rates and suboptimal outcomes.

Although high-dose intravenous penicillin remains the standard of care, our patient received broad-spectrum antibiotics. This

Table I. Overview of pediatric osteomyelitis cases attributed to *Actinomyces*: clinical presentation, diagnosis, and management.

Author (year)	Age (yr) / sex	Location	Histopathologic diagnostic method	Culture result	Antibiotic treatment
Chew et al.[6] (2023)	9 / F	Cervical spine	Histopathologic diagnosis	Anaerobic culture negative	Ampicillin → AMC
Chew et al.[6] (2023)	9/ F	Mandible	Excision debridement	Anaerobic culture negative	AMC
Chew et al.[6] (2023)	9/ F	Mandible	Incision and drainage of the abscess	*Actinomyces israelii*	AMC and ceftriaxone
Chew et al.[6] (2023)	12/ F	Mastoid	Incision and drainage, mastoidectomy	Anaerobic culture negative	AMC
Conley et al.[14] (2022)	17 / M	Anterior skull base	Medial orbitotomy for drainage	*Actinomyces*	Clindamycin, ceftriaxone
Iwai et al.[11] (2021)	14 / F	Parotid-masseter	Histopathologic diagnosis	Anaerobic culture not reported	Ampicillin, later amoxicillin
Mou et al.[12] (2021)	5 / F	Right lower extremity	Debridement and drainage	*Actinomycetes europaeus*	Ampicillin-sulbactam
Boorman et al.[9] (2020)	7 / M	Nasofrontal region	Sequestrum, debridement	*Actinomyces funkei*	Chloramphenicol
Saarinen et al.[16] (2011)	13 / M	Mandible	Debridement	*Actinomyces*	Penicillin
Saarinen et al.[16] (2011)	11 / F	Mandible	No	*Actinomyces turicencis*	Penicillin
Saarinen et al.[16] (2011)	5 / F	Mandible	No	*Actinomyces*	Penicillin
Saarinen et al.[16] (2011)	17 / F	Mandible	No	*Actinomyces*	Amoxicillin
Catalano-pons et al.[17] (2007)	6/ F	Iliac bone	Histopathologic diagnosis	*Actinomyces israelii*	Amoxicillin
Catalano-pons et al.[17] (2007)	7/ M	Elbow and the mandible	Histopathologic diagnosis	*Actinomyces*	Metronidazole and amphotericin B, followed by amoxicillin alone
Robinson et al.[15] (2005)	4/ F	Ramus and angle of the jaw	Debridement, sequestrectomy,	*Actinomyces israelii*	Amoxicillin-clavulanate
Robinson et al.[15] (2005)	3/ M	Mandible	Sequestrectomy	*Actinomyces naeslundii*	Penicillin → clindamycin, then amoxicillin
Sobol et al.[13] (2004)	14/ F	Temporal bone	Tympanomastoidectomy	Anaerobic culture not reported	Oral penicillin
Thisted et al.[19] (1987)	3/ M	Mandible	Puncture	*Actinomyces* spp.	Ampicillin, metronidazole
Vannier et al.[20] (1986)	13/ F	Skull and atlas	Puncture, drainage	*Actinomyces israelii*	Erythromycin and metronidazole

AMC: amoxicillin / clavulanic acid, F: female, M: male.

Table I. Continued.

Author (year)	Age (yr) / sex	Location	Histopathologic diagnostic method	Culture result	Antibiotic treatment
Walker, et al.[18] (1981)	7/ M	Mandible	Curettage	*Actinomyces israelii*	Penicillin
Present case	15/ M	Mandible	Drainage	Anaerobic culture not performed, aerobic culture negative	Vancomycin, metronidazole, cefotaxime → teicoplanin, piperacillin/tazobactam → meropenem, teicoplanin → at discharge: oral AMC

AMC: amoxicillin / clavulanic acid, F: female, M: male.

decision was based on the severity of the infection, the presence of mandibular osteomyelitis, and clinical instability at the time of histopathologic confirmation. Given these factors, de-escalation to penicillin was not pursued, and therapy was maintained with broad-spectrum coverage until clinical resolution. The literature consistently supports prolonged penicillin therapy as first-line treatment; however, tailored regimens may be necessary in severe, refractory, or complicated cases.

Imaging techniques, including CT and MRI, were essential for both diagnosis and monitoring.[22] These modalities help distinguish infection from neoplasm and track disease progression. On day 28, follow-up imaging demonstrated near-complete resolution. This aligns with literature emphasizing the value of serial imaging to guide treatment duration and detect complications.[22,23]

In conclusion, this case highlights the need for a multidisciplinary approach including infectious disease specialists, radiologists, otolaryngologists, microbiologists, and pathologists. Pediatric cervicofacial actinomycosis, though rare, should be considered in persistent infections with negative cultures and osteomyelitis. Multidisciplinary care improves diagnostic accuracy and treatment outcomes.

Ethical approval

A written informed consent was obtained from the parents of the patient for this publication.

Author contribution

The authors confirm contribution to the paper as follows: Case report conception and design: SYA, ST, MD, EY, OE; literature review: SYA; draft manuscript preparation: SYA, ST, MD, EY, OE. All authors reviewed the results and approved the final version of the manuscript.

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Conflict of interest

The authors declare that there is no conflict of interest.

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