

Case Report

A Solitary Skull Lesion of Syphilitic Osteomyelitis

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We experienced a rare case of solitary syphilitic osteomyelitis of the skull without any other clinical signs or symptoms of syphilis. A 20-year-old man was referred due to intermittent headache and mild tenderness at the right parietal area of the skull with a palpable coin-sized lesion of softened cortical bone. On radiological studies, the lesion was a radiolucent well enhanced mass (17 mm in diameter). The erythrocyte sedimentation rate (52 mm/h) and C-reactive protein (2.24 mg/dL) were elevated on admission. Serum venereal disease research laboratory (VDRL) and *Treponema pallidum* haemagglutination assay (TPHA) tests were positive. There were no clinical signs or symptoms of syphilis. After treatment with benzathine penicillin, we removed the lesion and performed cranioplasty. The pathologic finding of the skull lesion was fibrous proliferation with lymphoplasmocytic infiltration forming an osteolytic lesion. In addition, a spirochete was identified using the Warthin-starry stain. The polymerase chain reaction study showed a positive band for *Treponema pallidum*. Solitary osteomyelitis of the skull can be the initial presenting pathological lesion of syphilis.

KEY WORDS : Infectious osteomyelitis · Syphilis · Skull · *Treponema pallidum*.

INTRODUCTION

The history of syphilitic diseases⁷⁾, includes syphilitic osteomyelitis of the skull as a common finding³⁾. However, there are only few reports of a solitary skull lesion due to syphilitic osteomyelitis without any other clinical manifestations of *Treponema pallidum* infection. We report a case of early syphilis presenting with syphilitic osteomyelitis of the skull that was the only pathological lesion identified in the patient.

CASE REPORT

A 20-year-old man was referred to our hospital due to intermittent headache and mild tenderness of a small coin-sized and slightly bulging lesion with softened cortical bone at the right parietal skull area. A small round radiolucent lesion was found on plain skull X-ray (Fig. 1A). The com-

puted tomography (CT) and magnetic resonance imaging (MRI) revealed a 1.7 cm-sized, osteolytic and well enhanced lesion involving the whole thickness of skull bone; a solitary bone tumor was suspected (Fig. 1B, C). Bone scintigraphy demonstrated hot-uptake at the lesion (Fig. 1D). Preoperative laboratory tests showed evidence of syphilis; the erythrocyte sedimentation rate was 52 mm/h, the C-reactive protein (CRP) was 2.24 mg/dL, the venereal disease research laboratory (VDRL) test was positive, and *Treponema pallidum* haemagglutination assay (TPHA) was above 5,120 IU/L. However, the patient did not have any signs or symptoms of syphilis. The serology for the human immunodeficiency virus (HIV) was negative. There was no evidence of congenital syphilis or familial history of venereal diseases. The patient had a sexual encounter with a prostitute, which was the first and only sexual activity reported, 7 months previously. After three weeks of treatment with benzathine penicillin (intramuscular injection, 2,400,000 IU once a week), a resection of the lesion and cranioplasty with bone cement were performed simultaneously.

Hematoxylin and eosin staining showed fibrous proliferation with lymphoplasmocytic infiltration forming an osteolytic defect suggesting non-specific chronic inflammatory changes. A spirochete was identified with the Warthin-starry

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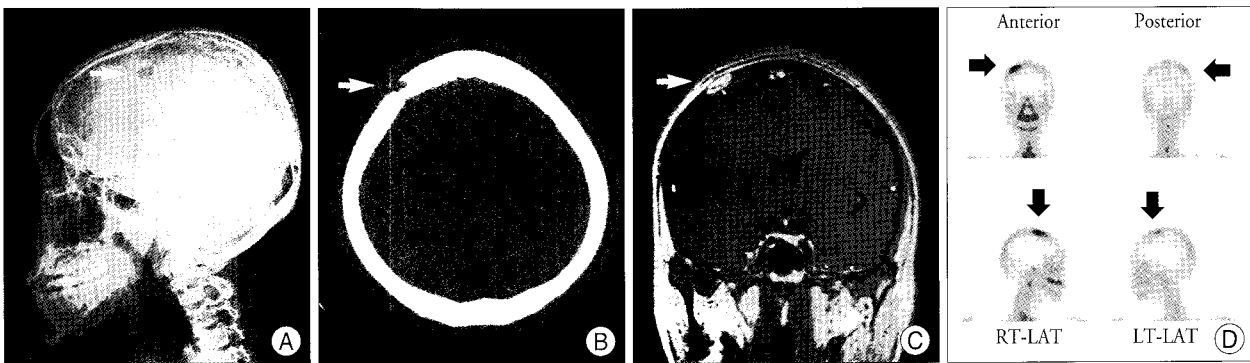


Fig. 1. The Results of the imaging study. A : Skull lateral x-ray shows a 17 mm radiolucent skull lesion (arrow) of parietal bone. B : Computer tomography shows a 17 mm osteolytic skull lesion (arrow) involving whole thickness of the right parietal bone. C : A coronal section of magnetic resonance imaging shows a well enhanced mass lesion involving the whole thickness of skull bone (arrow) at the right parietal area and surrounding soft tissue swelling. D : Bone scintigraphy shows a solitary hot uptake lesion (arrows) of the right skull.

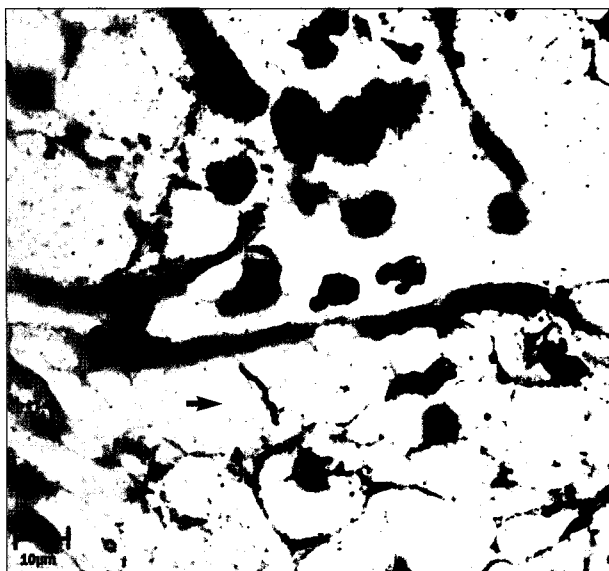


Fig. 2. Identification of spirochetes by Warthin-starry staining ($\times 1,000$). Arrow indicates the *Treponema pallidum*.

staining (Fig. 2). The result of the polymerase chain reaction (PCR) with skull pathology showed a positive band for *Treponema pallidum*¹. Cerebrospinal fluid (CSF) examinations, performed after confirming syphilitic osteomyelitis, were positive for VDRL testing without any evidence of inflammation.

DISCUSSION

Although there was no evidence by serologic test or pathological evaluations, *T. pallidum* infection is reported to have existed prior to AD 300⁷. Syphilitic osteomyelitis of the skull is a destructive bone disease that occurs as a complication of syphilis; however, it is an unusual complication of early stage (primary or secondary) syphilis^{9,10,14}. In this case, the patient presented only with a syphilitic osteomyelitis of the skull even though he had early stage syphilis according to his

clinical, familial, and social histories.

Recent epidemiological data from the US National Institutes of Health showed increasing prevalence of syphilis after 2002, and osteomyelitis lesions of the skull in early stage syphilis have been reported in HIV infected patients^{2,6,9,10}. The situation is similar in the Republic of Korea¹². Just like tuberculosis is increasing in HIV infected patients; syphilis also has been on the rise^{3,13,15}. There have been many reports of syphilitic osteomyelitis not associated with HIV, and likely many unreported cases^{4,7,11}. Syphilitic osteomyelitis can involve various bones including skull, clavicle, tibia, humerus, ulna, and the radius⁴. Syphilitic osteomyelitis can be treated with antibiotic therapy, benzathine penicillin or ceftriaxone^{4,16}. We performed surgery on this patient with the initial impression of a skull tumor; osteomyelitis of skull as the sole and initial presenting finding of syphilis is very rare in patients without an immunological disorder, especially in this country. Radiologically, it is difficult to distinguish a syphilitic osteomyelitis from other radiolucent lesions of the skull, such as, eosinophilic granuloma, multiple myeloma, or cystic fibrous dysplasia⁸. This case illustrates that syphilis should be included in the differential diagnosis and that a bone biopsy for pathological examination and PCR and/or VDRL test should be performed.

CONCLUSION

Syphilitic osteomyelitis of the skull as the sole initial presenting lesion is very rare, and can be diagnosed by identification of spirochetes in the lesion combined with a positive PCR or VDRL of affected bone.

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