Syphilis of the lumbar spine
A case report and review of the literature
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Abstract
Rationale: Tertiary syphilis can manifest as gummatous disease, but gumma of the spine has been extremely rarely reported.
Patient concerns: A 61-year-old male complained of worsening pain and numbness in both lower legs for four weeks.
Diagnoses: Syphilis of the Lumbar Spine.
Interventions: Pedicle screw fixation (L3-S1) and posterior decompression of the vertebral canal at L4-5 were performed.
Outcomes: The postoperativeVAS score of both lower extremities decline to 2 from 7 at admission. Dorsal thumb extensor motor power (left/right) at day 7 postoperatively was 3/3 (versus admission: 1/1). Laboratory examinations showed normal white blood cell count (versus admission: 13.8 × 10^9/L; reference value: 4.00–10.00 × 10^9/L) and decline in C-reactive protein (20.35 mg/L versus admission: 77.43 mg/L; reference value: 0.00–10.00 mg/mL) and ESR (58 mm versus admission: 73 mm; reference value: 0–15 mm).
Lessons: Our case illustrates that although gummatous disease of the spine may be extremely rare, it should be considered in the differential diagnosis of tuberculous or malignancy of the spine so as to avoid a wrong diagnosis and incorrect treatment.

Abbreviations: MRI = magnetic resonance imaging, VAS = visual analog scale.
Keywords: gumma, lesionectomy, lumbar spine, syphilis, vertebral decompression

1. Introduction
Syphilis is a sexually transmitted disease due to infection by Treponema pallidum subspecies pallidum and may present as a primary, secondary, latent, and tertiary, or congenital disease. The disease has gained clinical significance in China as it now ranks among the top 5 reported communicable diseases in the country after dramatic increase in prevalence over the previous decades.[1,2] The manifestations of late syphilis are rarely seen today, particularly given the availability of penicillin.

Tertiary syphilis manifests as gummatous disease. When gumma is formed in the spine cord, the imaging characteristics and some of clinical symptoms were in a high similarity with spine tuberculosis or spine tumors.[3,4] Therefore, the differential diagnosis is crucial. In this paper, we described a case of syphilis in a 61-year old man with syphilitic lumbar spine lesions and also carried out a literature review.

2. Case report
A 61-year-old male presented to our clinic with worsening pain and numbness in both lower legs for 4 weeks. The symptoms started 1 year ago, and difficulty in walking appeared 6 months ago. He denied having fever, chills, nausea, vomiting, cough, difficulty breathing, or arthritis. The patient had a 2-year history of syphilis and had been treated with penicillin for 14 months. The patient was sexually active but did not has a stable sexual partner. No history of other infections or malignancy was reported.

Physical examination revealed normal vital signs. No rashes, papules, or ulcers were found and there was no generalized lymphadenopathy. The patient had a VAS score of 7 of both lower extremities. Motor power (left/right) was graded as follows: quadriceps (4/4), tibialis anterior (3/3), and dorsal thumb extensor (1/1). No bowel dysfunction was detected. There were no oral or genital lesions. Laboratory investigations on admission showed mild anemia (hemoglobin 11.7 g/dL; reference value: 12–16 g/dL), increased white blood cell count (13.8 × 10^9/L; reference value: 4.00–10.00 × 10^9/L), elevated C-reactive protein (77.43 mg/L; reference value: 0.00–10.00 mg/mL), procalcitonin (1.18 μg/L; reference value: 0.00–0.05 μg/L), and ESR (73 mm; reference value: 0–15 mm). Serological tests for hepatitis B virus and hepatitis C virus were negative. Syphilis test results were as follows: RPR (+) and RPR 1:2 (+) HIV and Mantoux tuberculin skin test were negative.

Lumbar spine x-ray revealed loss of disc height and abnormal high density of the vertebral body at L4 and L5 (Fig. 1). Whole
spine magnetic resonance imaging (MRI) identified severe narrowing of the disc space at L4–5 and a mass of 4.3 mm × 1.8 mm × 1.6 mm in size, which was located immediately behind the vertebral body at L4 (Fig. 2). The mass had a sharp boundary. A lumbar CT scan showed extensive wormy appearance throughout the lumbar spine, which was the most noticeable at L4 and L5 (Fig. 3).

Posterior decompression of the vertebral canal at L4–5 was performed after pedicle screw fixation (L3-S1) (Fig. 4). The dural sac was pulled to the left side and the mass was found under the posterior longitudinal ligament and tenaciously attached to adjacent tissues. The mass was white–gray, rubbery, and resembled a tumor mass. Nucleus pulposus clamp and bone curets were used to remove the mass from the vertebral body at L4. There was no massive bleeding during the procedure and the mass appeared white–gray and was rubber (Fig. 5). The postoperative VAS score of both lower extremities was 2. Dorsal thumb extensor motor power (left/right) at day 7 postoperatively was 3/3. Laboratory examinations showed normal white blood cell count and decline in C-reactive protein (20.35 mg/L) and ESR (58 mm). Pathological slices showed degenerative necrotic tissues and fibrous connective tissues with occasional perivascular infiltration by lymphocytes (Fig. 6).

3. Discussion

Gummatous disease of the spine has rarely been reported. Here, we describe a case of syphilis in a 61-year old male who developed gummatous disease of the spine 2 years after his initial diagnosis of syphilis. Early studies suggested that syphilitic bone involvement may be expected in approximately 1 to 5% of untreated syphilis patients,[5,6] but the prevalence of gummatous disease of the spine remains unknown as this particular type of complication has only been anecdotally reported. Yang et al[4] have recently reviewed the literature published in PubMed and Embase searches between 1960 and 2015 and identified 8 cases (including 1 case of their own) of spinal syphilitic gumma. They included 5/8 females with age ranging from 25 to 65 years of age. The clinical manifestations included paraplegia, sensory disturbance, and back pain. Six out of eight patients underwent lesionectomy. Our patient also had sensory disturbance and lower leg pain. One noticeable feature of the patient is that the gummatous disease of the spine had developed despite penicillin treatment for 12 months. Posterior decompression of the vertebral canal and lesionectomy were performed in the patient, which improved motor function and reduced leg pain. Reported cases by Yang et al[4] included a 38-year old male with syphilis of the spine for 1 year, who was treated with the decompressing laminectomy of the fourth lumbar vertebra.[5] Johns[3] also reported 2 cases of syphilis of the spine, involving the sixth and seventh thoracic vertebra in a 67-year old female and the fourth and fifth lumbar vertebra in another 51-year old woman. Grifins and Rose[7] described a patient with a history of syphilis for 20 years who developed cauda equina compression. Laminectomy of the fourth and fifth lumbar vertebrae was performed. Though no mass was found in the extradural tissues, a cavity was detected in the position of the fifth lumbar vertebral body.

Gummatous disease of the spine may simulate tuberculosis infection and malignancy of the spine. Two cases reported by Johns[3] were initially mistaken for tuberculosis of the spine while the case by Yang et al[4] was first mistaken for spinal intramedullary tumor. The diagnosis of gummatous disease of
Figure 2. Spinal magnetic resonance imaging (MRI) identifies severe narrowing of the disc space at L4–5 and a mass 4.3 mm × 1.8 mm × 1.6 mm in size immediately behind the vertebral body at L4. The mass shows mixed signals on both T1 and T2-weighted imaging (C and D). In the axial image, the mass occupies 70% of the vertebral canal (B).

Figure 3. Lumbar CT scan shows extensive wormy appearance throughout the lumbar spine, which is the most noticeable at L4 and L5. (A) the L3 vertebra; (B) the L4 vertebra; (C) the lower L4 endplate; (D) the upper L5 endplate. CT = computed tomography.
the spine in our case was hinted by the positive syphilis test result and by intraoperative pathological findings showing the gumma contained a center of coagulated necrotic material and margins consisting of palisaded fibroblasts and macrophages and mononuclear cells. Furthermore, Mantoux tuberculin skin test was negative in the patient, ruling out the likelihood of tuberculous infection of the spine.

Given the rise in the incidence of syphilis in China, the diagnosis and treatment of syphilitic disorder of the spine may once again become a significant problem in orthopedic surgery. Although gummatous disease of the spine may be extremely rare, it should be considered in the differential diagnosis of tuberculosis or malignancy of the spine to avoid a wrong diagnosis and incorrect treatment.

Figure 4. Lumbar spine x-ray shows posterior decompression of the vertebral canal at L4–5 canal after pedicle screw fixation (L3–S1). (A) The anteroposterior view; (B) the lateral view.

Figure 5. Photograph of the intraspinal mass appeared. It is white–gray and rubbery.
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Figure 6. (A) Hematoxylin & eosin (H&E) staining of the resected tissue is consistent with syphilitic granuloma (original magnification, ×20). (B and C) Photomicrograph shows fragments of necrotic bone embedded in fibrous tissue (H&E stain; original magnification, B ×40 and C ×100).