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Mononeuritis Multiplex as the Initial Manifestation of Candida Infective Endocarditis

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Study Design: Case report.
Objectives: To report a case of mononeuritis multiplex as the initial manifestation of Candida infective endocarditis (IE).
Summary of Literature Review: Mononeuritis multiplex is actually a group of diseases, not a distinct disease entity. It often results from blood vessel diseases, diabetes, or inflammation due to autoimmune connective tissue disorders, although many cases are idiopathic. IE is an infection of the endocardial surface of the heart. Multiple nerves were affected simultaneously in several cases of IE, making consideration of IE important in the differential diagnosis of mononeuritis multiplex.
Materials and Methods: We present a rare case of a 71-year-old man with IE in whom mononeuritis multiplex was revealed on electromyography; further, he presented with lower back pain and sciatica.
Results: The presence of the characteristic symptoms of lumbar radiculopathy in this case delayed the diagnosis and proper treatment of the patient.
Conclusions: Physicians should carefully consider all patient-related data, and also provide accurate information to consultants when they refer patients. This can help to prevent serious complications.

Key words: Mononeuritis multiplex, Infective endocarditis, Lumbar radiculopathy

Introduction

Mononeuritis multiplex is a painful asynchronous sensory and motor peripheral neuropathy involving isolated damage to at least 2 separate nerve areas.[1] This somewhat exotic term has been used to describe the classical and most frequent pattern of vasculitic neuropathy.[2] The physiological and neurological examinations, and past history, of patients with suspected vasculitis are broad and multi-systemic. As a result, one of the most frequent pitfalls in the diagnosis of these diseases is restricting the clinical evaluation of a patient to one department or the expertise of only the consulting clinician.[2,3]

Infective endocarditis (IE) is an infection of the endocardium of the heart. Despite IE being an uncommon diagnosis for a generalist, it may nonetheless present with a wide diversity of, sometimes subtle, clinical signs; diagnosis may be difficult or the signs misleading, and there are wide differential diagnoses to consider. IE is also one of the differential diagnoses of mononeuritis multiplex that physicians should recognize.[4] Although many microorganisms can cause IE, Candida IE, despite being uncommon, is often lethal.[5] Here, we present a rare case of a 71-year-old man with IE in whom mononeuritis multiplex was revealed on electromyography; further, he presented with lower back pain and sciatica. The presence of the characteristic symptoms of lumbar radiculopathy in this case delayed the diagnosis and proper treatment of the patient.

Case Report

A 71-year-old man was transferred to our hospital with a 3-week history of severe pain in the lower back and right leg, along the L5–S1 dermatome, and excessive fatigue, which

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started after he received treatment for the common cold. He complained of having a sensation of heat and sometimes broke out in a cold sweat on his lower back without fever or chill. His medical and surgical history was unremarkable. There was severe pain in his right leg radiating from the buttocks to the calf and he showed weakness in his right foot plantar flexion. A straight-leg raise test was positive at 50° on the right side. Prior to presentation, the patient had undergone lumbar magnetic resonance imaging (MRI) at another hospital; the MRI revealed no significant findings (Fig. 1). Nevertheless, he was treated with oral antibiotics and non-steroidal anti-inflammatory drugs under a working diagnosis of pyogenic spondylitis because laboratory investigations at another hospital revealed a 10-day history of increased erythrocyte sedimentation rate (ESR) and C-reactive protein (CRP) levels and severe pain on lower back. However, at the time of transfer to our hospital, his pain was more aggressive. On admission, laboratory investigations revealed an ESR of 24 mm/h (normal range: 0–20 mm/h) and a CRP concentration of 50.6 mg/L (normal range: 0–5 mg/L), white blood cells count of 4910/μl (normal range; 4,000-10,000/μl), hemoglobin of 9.5 g/dL (normal range: 13-18 g/dL), and a platelet count of 61,000/μl (normal range; 150,000-450,000/μl). A peripheral blood smear examination revealed normocytic normochromic anemia with anisopoikilocytosis (annulocyte and elliptocyte) and thrombocytopenia. A three-phase bone scan revealed no significant infection signs except for polyarticular joint disease involving the large and small joints of both upper and lower limbs, and the sacroiliac joints. Electromyography (EMG) and nerve conduction velocity (NCV) tests revealed right-sided sciatic neuropathy with mononeuritis multiplex patterns suggesting vasculitic neuropathy. Seven days after admission, we observed pitting edema on both his legs. His blood culture test was positive for Candida parapsilosis. Transthoracic echocardiogram (ECG) revealed vegetation on the aortic valve (24×18 mm) and moderate aortic stenosis with mild aortic regurgitation due to the vegetation: highly indicative of IE (Fig. 2). We recommended a transfer to the department of thoracic and cardiovascular surgery and cardiology for treatment; his family wanted the transfer to be to a hospital near their hometown. He underwent aortic valve replacement surgery at that hospital and died 10 days after operation due to surgical complications.

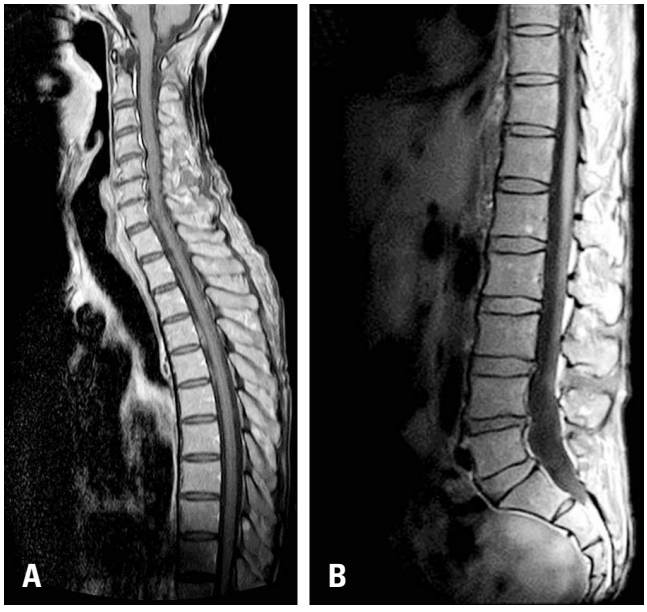

Fig. 1. Gadolinium-enhanced sagittal magnetic resonance imaging **(A , B)** showing no significant abnormality to explain the pyogenic spondylitis.

Discussion

Mononeuritis multiplex is actually a group of diseases, not a distinct disease entity.[6] It demonstrates the sequential involvement of individual nerves or trunks usually in a distal to proximal fashion, and an asymmetrical pattern.[2] Multiple nerves in seemingly-random parts of the body can be involved. As the condition worsens, the symptoms become less multifocal and more symmetrical. Mononeuropathy multiplex syndromes can be bilaterally, distally, or proximally distributed.[6] An intimate

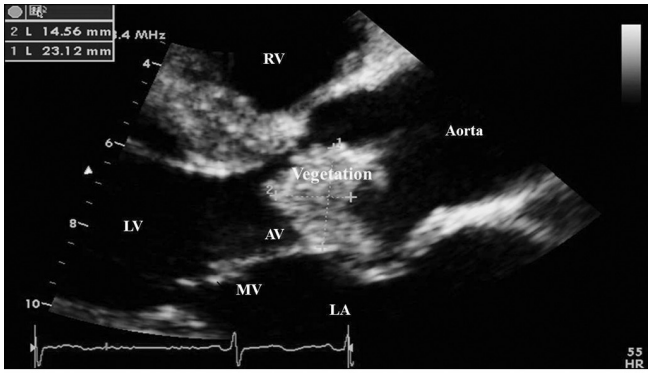

Fig. 2. Transthoracic echocardiogram showing vegetation on the aortic valve (24 × 18 mm) and moderate aortic stenosis with mild aortic regurgitation due to vegetation: highly suggestive of infectious endocarditis. AV = aortic valve; LA = left atrium; LV = left ventricle, MV = mitral valve; RA = right ventricle.

and comprehensive medical history taking is thus very important in identifying its possible underlying cause. The pain often starts in the low back or hip and extends to the thigh and knee on one side. The pain is most commonly deep and aching, with superimposed lancinating jabs that are most severe at night. Patients with diabetes typically complain of an acute onset severe unilateral thigh pain that is followed rapidly by weakness and atrophy of the anterior thigh muscles and loss of the knee reflex.[6]

The possible cause of mononeuritis multiplex, as suspected by the medical history, symptoms, and pattern of symptom development of the patient, helps to determine which tests to perform.[2] Mononeuritis multiplex often results from blood vessel diseases, diabetes, or inflammation due to autoimmune connective tissue disorders, although many cases are idiopathic. Therefore, a number of consultations could be useful in the treatment of mononeuritis multiplex: 1) neurologists − if an underlying neurologic condition is suspected, 2) rheumatologists − if an underlying rheumatologic condition is suspected, 3) infectious disease specialists − if evidence of an infectious etiology is present, 4) pain management specialists, or physiatrist referrals, may be needed in selected cases.[6] The physician must try to detect the underlying cause and begin the proper treatment according to the established protocols for the specific disease condition. Some diseases can be lethal if not treated properly.[3]

IE is an infection of the endocardial surface of the heart, which may include one or more heart valves, the mural endocardium, or a septal defect.[4] If untreated, IE is often fatal.[7] Staphylococci and streptococci account for 80% of cases of IE, with staphylococci currently the most common pathogen.[4] Musculoskeletal symptoms such as arthralgia, myalgia, and back pain, are common manifestations during IE, and sometimes rheumatic problems may be the first symptoms of the IE.[3] Griffin reported that IE was diagnosed in 30.8% of patients with pyogenic spondylodiscitis and was more common in cases of streptococcal infection and predisposing heart conditions.[3] When IE is suspected, transthoracic ECG should be performed as soon as possible.[7] IE patients with back pain should undergo MRI or computed tomography of the spine. Conversely, transthoracic ECG may be performed in patients with a definite diagnosis of pyogenic spondylodiscitis and underlying cardiac conditions predisposing the patient to endocarditis.[3]

Acute cranial and peripheral mononeuropathies have been reported largely in IE caused by viridans streptococci.[8] Cutaneous emboli and splinter hemorrhages corresponded to the peripheral nerve involved, and embolic occlusion of the vasa vasorum is a more likely mechanism than immune−mediated injury in the early phase of the disease. Multiple nerves were affected simultaneously in several cases, making consideration of IE important in the differential diagnosis of mononeuritis multiplex.[8] Fungi are an uncommon cause of IE and account for only 1 − 6% of total cases. Candida species are the most common causes of fungal endocarditis and the overall mortality rate of Candida IE is more than 50%, despite treatment.[5] Although the epidemiology, risk factors, and outcomes of Candida IE are not well known, due to its rarity and the lack of large prospective cohort studies, the clinical findings and presentation of patients with Candida and non−fungal IE are very similar.[4] The survival rate of fungal endocarditis has increased over the past twenty years, from 14% before 1970 to 41% in 1991 − 1995.[9] Better ECG techniques, rapid diagnosis, and appropriate supportive care of ill patients are the factors predominantly causing the increased survival rates.[9] Treatment necessitates dual antifungal administration and valve replacement. Most cases are treated with various forms of amphotericin B with or without azoles, although recent case reports describe successful therapy with the new echinocandin caspofungin.[10]

Conclusion

Our patient was treated for 10 days with suspected pyogenic causes, because of his increased ESR and CRP, and his spinal symptoms even without abnormalities on his lumbar MRI. After he was transferred to our hospital, we did not identify the symptoms, and pattern of symptom development of the patient because we were focusing on his spinal problem; for similar reasons, his medical history was insufficiently detailed/complete. Consequently, accurate diagnosis of the patient was delayed for 17 days, despite his needing rapid and proper treatment. A retrospective review of this patient revealed that all his symptoms and previous examinations were consistent with IE. Therefore, a definitive diagnosis could have been obtained in a short time. Physicians should carefully consider all patient−related data, and also provide accurate information to consultants when they refer patients. This can help to prevent serious complications.

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Case Report

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칸디다 감염성 심내막염 초기증상으로 나타난 다발성 단일신경염

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연구 계획: 증례보고
목적: 칸디다 감염성 심내막염 초기증상으로 나타난 다발성 단일신경염 1예를 보고한다.
선행문헌의 요약: 다발성 단일신경염은 실제로 여러 질환들의 그룹으로 분명한 질병단위는 아니며 혈관 질환, 당뇨, 자가면역성 결합조직 질환들에서 발생할 수 있으나 많은 경우에서 원인 불명으로 알려져 있다. 감염성 심내막염은 심장내막 표면의 감염으로 동시에 다발성 신경들이 영향을 받기도 하므로 다발성 단일신경염의 감별진단에 있어 중요하게 고려된다.
대상 및 방법: 요통 및 하지 방사통으로 전원되어 시행한 근전도 및 신경전도검사상 다발성 단일신경염을 보인 71세 남자에서 감염성 심내막염으로 진단된 증례를 보고한다.
결과: 신경근병증의 특징적인 증상으로 인해 진단 및 적절한 치료가 지연되었다.
결론: 신속하고 적절한 치료를 위해 환자와 관련된 모든 데이터를 주의 깊게 확인해야 한다. 또한자문의에게 정확한 정보 제공을 위해 노력 해야 하며 이는 심각한 합병증을 막는데 도움이 될 것이다.

색인 단어: 다발성 단일신경염, 감염성 심내막염, 요추 신경근병증
약칭 제목: 칸디다 감염성 심내막염에서의 다발성단일신경염

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