Case Report
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Sepsis-like presentation of giant cell arteritis

Abstract

Introduction: Giant cell arteritis (GCA) is a subacute inflammatory panarteritis affecting the elderly. The clinical manifestations of this vasculitis are often progressive, combining cephalic, general, and systemic signs. A sudden onset mimicking an infectious disease may occur and should not lead to misdiagnosis in the elderly. Pseudo-septicemic forms are exceptional. We report an original and unusual sepsis-like presentation of GCA.

Case Report: A 66-year-old woman, with no significant past medical history, was hospitalized for a fever of 39-40°C that had been present for 15 days. Her general condition was good, and the physical examination was unremarkable. Laboratory tests revealed a marked inflammatory syndrome with hyperleukocytosis. The search for direct or indirect signs of infection was negative, and broad-spectrum antibiotic therapy did not improve the patient's condition. A temporal artery biopsy was performed and confirmed the diagnosis of GCA. Systemic corticosteroid therapy was initiated, and the patient's condition improved rapidly.

Conclusion: GCA can be mono- or pauci-symptomatic, presenting only as rheumatic, ocular, neuropsychiatric, pulmonary, neoplastic-like, sepsis-like, or biological manifestations. These atypical forms should not delay diagnosis in order to avoid serious ischemic complications of this disease.

Keywords: giant cell arteritis, sepsis-like, horton's disease, sepsis, polymyalgia arteritica, vasculitis

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Introduction

Giant cell arteritis (GCA), also known as polymyalgia arteritica, temporal arteritis, and Horton's disease, is a primary subacute inflammatory panarteritis of large vessels affecting classically the elderly over 50 years.[1,2] The clinical manifestations of this vasculitis are often progressive, combining cephalic involvement, nonspecific general symptoms, and systemic signs.[1,3] Fever is observed in more than half of cases of this disease and is most often moderate.[1,3] but a sudden onset mimicking an infectious disease may occur and should not lead to misdiagnosis in the elderly.[4] Pseudosepticemic or sepsis-like presentation of this disease is exceptional and presents a real diagnostic challenge for clinicians, particularly in the elderly where infections are frequent.[5] Furthermore, the infectious risk of the disease itself is increased, making it difficult to distinguish between a true, sometimes serious, infectious complication of the disease and a simple pseudo-infectious presentation.[6,7] We report an original and unusual presentation of GCA mimicking sepsis in a 66-year-old Tunisian woman.

Case presentation

A 66-year-old Tunisian woman, with no significant past medical history, was hospitalized for a fever of 39-40°C that had been present for 15 days. She reported a dry cough for which she had been treated with a quinolone with no improvement. Her general condition was good, and the physical examination showed no significant abnormalities other than isolated fever of 40°C.

Laboratory tests revealed a marked inflammatory syndrome with a C-reactive protein (CRP) at 40 mg/L, an erythrocyte sedimentation rate (ESR) at 120 mmH1, and polyclonal hypergammaglobulinemia at 24g/L on plasma protein electrophoresis. The blood count showed neutrophilic hyperleukocytosis at 15,000/mm³ with 80% neutrophils and normochromic normocytic anemia with hemoglobin at 10 g/dL. Other basic laboratory tests were normal (glucose, creatinine, uric acid, lipid profile, transaminases, thyroid function, calcium, and urinalysis). Chest X-rays was normal, immunological tests were negative, transesophageal echocardiography was normal, tumor markers were negative, and thoraco-abdomino-pelvic CT scan showed no significant abnormalities.

The search for direct or indirect signs of infection was negative (blood cultures, urine culture, sputum culture, various bacterial, viral, parasitic and fungal serological tests, cerebrospinal fluid analysis, Quanteferon, and tuberculosis tests), and broad-spectrum antibiotherapy did not improve the patient's condition. A temporal artery biopsy was performed and confirmed the diagnosis of GCA showing granulomatous vasculitis with giant cells and fragmented internal elastic lamina. Systemic corticosteroid therapy at a dose of 0.7 mg/kg/day was initiated, and the patient's condition improved rapidly, with resolution of fever after three days and normalization of the CRP and leukocyte count after ten days.

Discussion

GCA is the most common vasculitis in the elderly.[1–3] Its frequency is largely estimated according to ethnic groups and countries with a notable South-North gradient: 0.34 cases per 100,000 people over 50 years old in East Asia to 21.57 cases per 100,000 people over 50 years old in Scandinavian countries.[1–3] Its clinical manifestations are by far dominated by headaches, polymyalgia rheumatica, ocular involvement and ischemic vascular complications affecting the skin, brain and abdominal viscera.[1,3,4] Rarely, GCA can present with several unusual and unexpected manifestations, making its diagnosis a real challenge for clinicians.[8–10]

Fever, a classic and frequent sign of GCA,[1,3,4] may remain the only manifestation of the disease,[11,12] making diagnosis even more challenging for clinicians, and GCA is the most frequent specific cause of prolonged fevers of unknown origin.[13] Fever during GCA is usually moderate, persistent, and prolonged.[1,3,4] Acute forms

mimicking severe infection or sepsis (pseudosepticemic or sepsis-like) are exceptional and unusual, with only a few sporadic cases reported in the world literature.[5,14,15]

These presentations are particularly difficult to diagnose in the elderly due to the increased risk of infection in this population,[5] as well as the well-documented risk of infection specific to GCA.[6,7] Indeed, in the series by Durand M et al of 1,664 patients with GCA, the adjusted rate ratios for systemic serious infections during the follow-up was 1.55, significantly higher than that of the matched control group of patients without GCA ($P < 0.001$), and the rate ratio for sepsis was 1.63.[6]. Similarly, Singh JA et al, in their study, observed a significantly higher sepsis rate in patients with GCA compared to the general population: 18.9% vs. 10.2%.[7]

In these cases, it is very difficult to distinguish a sepsis-like presentation of GCA from true sepsis or bacteremia that can mimic GCA.[5,14–16] Lack of response to antibiotics, spectacular improvement with systemic corticosteroid, as well as fluorodeoxyglucose positron emission tomography computerized tomography (18-FDG PET scan) imaging, and temporal artery biopsy would be very helpful in supporting the correct diagnosis.[5,14,15]

Conclusion

Giant cell arteritis can be mono- or pauci-symptomatic, presenting as only rheumatic, neuropsychiatric, ocular, pulmonary, neoplastic-like, sepsis-like, or biological manifestations. Pseudosepticemic or sepsis-like presentation of GCA, as our case report, is exceptional and uncommon. These atypical forms deserve to be known by healthcare professionals in order to avoid serious ischemic complications of this disease.

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Conflicts of interest

The author declares that there are no conflicts of interest.

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