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Case Report

Vascular Involvement in Behcet's Disease: A Case Report

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Abstract

A 68-year-old male with a 20 years long history of hip stiffness, acne like spots of trunk, oral ulcerations (aphthae), lower limb dermatitis and pericarditis presented to our hospital. He has been previously treated for arthralgia, periodic fever, and follicular lesions of the trunk in in- and out-patient settings. Imaging tests with Computed Tomography (CT) scanning showed inflammation of the abdominal aorta and colonoscopy revealed ischemic lesions of the intestines. A diagnosis of Behcet Disease (BD) with aortic involvement was established and a treatment scheme with Disease-Modifying Anti-Rheumatic Drugs (DMARDS), anti-TNF, anti-IL-6, and low dose colchicine was initiated. Currently, the patient was treated with injectable Canakinumab (Anti-IL1b), with excellent response.

Keywords: Behcet's disease, Canakinumab, Aortitis, Autoimmune diseases

Introduction

Behçet's Disease (BD), also known as Behcet's syndrome, is a rare auto-inflammatory disease that mainly affects the skin, joints, and mucus membranes [1]. Less commonly it affects the ocular central nervous system and large vessels, such as the aorta [2]. In this case, aortitis developed during the progression of the disease is considered an uncommon complication.

Case Report

A 68-year-old male patient presented to our clinic due to chronic fever (afternoon flares up to 38°C. for more than 15 years), arthralgia and easy fatigue. He has visited several hospitals, was subjected to a number of investigations and treatments that did not lead to a definite diagnosis and notable alleviation of the symptoms. The patient indicated the onset of the symptoms during childhood, with the appearance of periodic fever (lasting 3 days), exudation of follicular lesions of the trunk, and pain in joints. Laboratory and imaging investigation for infections and malignancies were negative at that time. Symptoms subsided after taking NSAIDs and corticosteroids.

He remained asymptomatic until the age of 20, when he developed:

a. daily fever up to 37°C,

b. upper and limb arthritis,

c. erythematous lesions of the upper extremities.

d. pericarditis

e. negative immunological control and high Inflammation.

After multiple hospitalizations and outpatient consultations in rheumatology clinics, RA with systemic manifestations was diagnosed. Following this diagnosis, he was initially treated with cor-

tisone and then with DMARDS and biological agents, without response (remission of pericarditis only). He was presented in our hospital with

a. hip stiffness, without peripheral arthritis,

b. acne like spots the trunk, aphthae.

c. lower-limb dermatitis.

d. high inflammatory markers.

Computed tomography of the abdomen was performed, and results showed: inflammation of the abdominal wall of the aorta and intestinal tract. Colonoscopy with biopsy showed: ischemia of the intestine without interferon cells. Negative cultures and serological control and the duration of the fever rule out infections. Negative gonadal control, daily fever and abdominal aortic involvement remove the case of autoimmune diseases. The presence of aortitis and the absence of inflammatory markers including ferritin, periods of remission ruled out Still disease.

There was administration of colchicine 2mgr/d, with remission of fever, and other symptoms but not of CRC, CRP. After 6 months our patient had partial remission but with intense abdominal pain. A Behçet's disease diagnosis was established, following the failure of treatment with cyclophosphamide. Finally, injectable canakinumab 300mgr/4w was administered, leading to complete remission to date.

Conclusion

Behçet's disease is relatively uncommon. It takes years for patients to meet the current diagnostic criteria and hence it can take years to reach such a diagnosis as happened in this case. This brings physical, mental, and financial burden to patients and healthcare systems, cultivates uncertainty, and undermines compliance to medical treatment and monitoring protocols. If a diagnosis is established, canakinumab is a very good alternative in resistant forms of the disease with aortic involvement. In this case, although Canakinumab is considered an off – label treatment, the patient responded well achieving a complete remission of aortitis to date.

Statement of Ethics

The patient has consented to the presentation of anonymized information related to their medical history.

Conflict of Interest

The authors have no conflict of interest to declare.

Acknowledgement

Not applicable.

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