

Case Report

Oro Genital Candidiasis Mimicking as Behcet's Syndrome: A Diagnostic Dilemma

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Abstract: Behcet's disease is a systemic inflammatory disease of unknown etiology. The disease is strongly associated with human leukocyte antigen (HLA B51). It has a chronic course with periodic exacerbations and progressive deterioration. There are no specific diagnostic laboratory tests, although recurrent oral ulceration is an obligatory manifestation for diagnosis. The treatment depends on severity of the illness and requires a multidisciplinary approach. This paper summarises a case presenting a diagnostic dilemma as it was initially thought to be Behcet's disease but later on it turned out to be a case of orogenital candidiasis.

Keywords: Behcet's disease, Orogenital candidiasis, Human leukocyte antigen, Recurrent aphthous ulcer

INTRODUCTION

Behcet's disease was originally described by a Turkish dermatologist Hulusi Behcet who noted a complex of recurrent oral and genital ulceration and iritis in a group of patients [1]. Environmental and genetic background may play a role as this disease is commonly found in the route that links Japan and Turkey otherwise known as silk route. The etiology is not well defined, but it is characterized by inflammation of blood vessel. The human leukocyte antigen HLA B51 which is linked to this disease is found commonly in the people of this region [2]. The triggering event for Behcet's disease has been debated for years. Possible etiologic agents indicate pollutant metals, English walnuts, organic insecticides and various microorganisms viz Streptococcus sanguis. M:F ratio is 2.1:1. The mean age of onset is 25-30 years. Due to ocular and CNS involvement in some reported cases it can lead to lethal complications and overall mortality rate is 16% in five years [3].

CASE REPORT

A 45 year old male presented to the Ophthalmology OPD of our tertiary care hospital with complaints of redness and profuse watering from both the eyes and diminished vision which was accompanied with pain. Patient was referred to Medicine OPD for general check up. On detailed history taking he revealed that initially he had developed redness and profuse watering from both the eyes three days back which was followed two

days later by widespread ulceration all over the oral cavity due to which he was unable to speak, swallow or open his mouth. Later on he also started complaining of burning micturition. Patient was admitted to our department for further evaluation and management for the above complaints. Patient also had history of oral ulceration one year back but was relieved by oral medication prescribed by a local doctor. No history of fever, joint pain, rashes, history of any drug intake, petechial haemorrhage or any immune compromised condition was present. Patient had history of tobacco chewing since last ten years. There was neither any history of immunosuppressive drug intake nor any immune compromised state i.e diabetes mellitus, chronic renal failure.

On local examination

Oral cavity

Widespread ulceration with reddish base was present extensively all over the buccal mucosa, palate, undersurface of the tongue, over the anterior two third of tongue, palate and both the upper and lower lip. Ulcers were painful and oral cavity was filled with slough and mouth opening was restricted (Fig. 1). Underlying oral candidiasis was present after removal of slough and debris.

Eye

Subconjunctival congestion with haemorrhage was present in both the eyes (Fig. 2). Bilateral fine keratic

precipitates was present at the back of cornea, grade II cells and grade I flare with synechiae and circum ciliary congestion present; pupil was dilated under drug. Distant vision was 6/9 in both the eyes. Bilateral fundus was within normal limit. Ophthalmology consultation was done and was diagnosed as a confirmed case of anterior uveitis.

Genitalia

A pseudo membrane was present all over the glans penis extending up to upper shaft of the penis except around the urethral meatus, a raw reddish area/ulcer was present. On skin consultation oral and genital candidiasis was confirmed (Fig. 3).

Rest of the general and systemic examination was within normal limit.

Routine investigations of the patient did not reveal any significant abnormality. ECG, chest X-ray PA view and other haematological reports were within normal limit. ELISA for HIV for the patient was non-reactive.

Patient was treated with IV antibiotics, IV fluids, corticosteroids, topical and oral antifungal agent was started, eye drops in the form of antibiotics with corticosteroids and cycloplegics, anti-inflammatory drugs and other symptomatic and supportive treatment was given. Patient recovered from his illness and was discharged.

Fig. 4-6 shows recovery one week after treatment of oral, genital and ocular lesions respectively. Subsequent follow up of the patient shows complete resolution from all the above sign and symptoms.



Fig. 1: Widespread Oral ulceration with slough



Fig. 2: Ocular involvement with subconjunctival haemorrhage in both the eyes



Fig. 3: Pseudo membrane extending over glans except over urethral meatus



Fig-4



Fig-5



Fig-6

Fig. 4-6: Resolution of Oro-genital-ocular lesion after one week treatment

DISCUSSION

There are no specific diagnostic tests for Behcet's disease. It is diagnosed clinically by specific patterns of symptoms and repeated outbreaks. Due to difficulty in establishing Behcet's disease the International study group for Behcet's disease formulated a set of criteria in 1993 [4]. These criteria require the presence of recurrent oral ulcerations at least thrice in one 12-month

period and at least 2 additional criteria, including recurrent genital ulcers, ocular lesions, skin lesions, and a positive pathergy test. Clinical diagnosis of Behcet's syndrome is based solely on diagnostic criteria set by International study group for the disease. The oral ulcerations may be minor aphthous ulcer, major aphthous ulcer or herpetiform. These criteria are based on the fact that more than 95% of cases had recurrent

aphthous ulceration. The additional criteria have been included in order to maximize the discrimination between Behcet's disease versus aphthous ulcer patients with another etiology viz orogenital candidiasis etc. The criteria states that a diagnosis of Behcet's disease requires recurrent oral ulceration and at least two additional criteria, including recurrent genital ulcers, ocular lesions, skin lesions and a positive pathergy test [5]. The genital ulcers are less common but are more specific, do not affect the glans penis or urethra, and produce scrotal scars on healing [6]. Behcet's disease is also linked to the presence of HLA B51 allele, this is more common in greek population [7]. Other systemic manifestations of Behcet's syndrome includes non deforming arthritis which involves knee and ankles, superficial or deep peripheral vein thrombosis, presence of aortitis or peripheral arterial aneurysm and thrombosis. Neurological involvement are less frequent but are reported (CNS-Behcet's syndrome) [8].

Orogenital candidiasis is a fungal infection (mycosis) caused by any species from the genus candida, most common being candida albicans [9] followed by candida tropicalis [10]. Candidiasis encompasses infections that range from superficial, such as oral thrush and vaginitis, balanitis to systemic and potentially life threatening diseases. A recently proposed classification of oral candidiasis distinguishes primary oral candidiasis where the condition is confined to mouth and perioral tissues and secondary oral candidiasis where there is involvement of other parts of body in addition to the mouth [11]. Candida infections can be of three types i.e pseudo membranous, erythematous (atrophic) and hyperplastic [12]. Acute pseudo membranous candidiasis is a classic form of oral candidiasis commonly referred to as thrush and accounts for 35% of all cases. Pseudo membranous candidiasis can involve any part of the mouth, but usually it appears on the tongue, buccal mucosa or palate [13]. Chronicity of this subtype generally occurs in immune compromised states e.g. leukaemia, HIV or in a person using corticosteroids topically or by aerosol [12]. Acute and chronic oro genital candidiasis are indistinguishable in appearance [14].

Chronic mucocutaneous candidiasis refers to a group of rare syndrome characterized by chronic candidal lesions on the skin, in the mouth and on other mucous membranes i.e. secondary oral candidiasis. These include localized chronic mucocutaneous candidiasis (candida granuloma, candida endocrinopathy). About 90% of people with chronic mucocutaneous candidiasis have candidiasis in mouth [14].

The above discussed case presented with recurrent aphthous ulceration, genital ulcers and above all what made the clinical picture more confusing was anterior uveitis. The patient responded well to antifungal treatment and did not fit in the diagnostic criteria of Behcet's syndrome. Anterior uveitis could be an

incidental finding in our case which created a confusing picture and made us to clinically suspect the above to be a case of Behcet's disease.

CONCLUSION

There is no confirmatory test for Behcet's syndrome. Diagnosis is solely based on clinical picture and certain criteria set by the International committee on Behcet's disease. Whenever a patient presents with orogenital lesions and ophthalmic features as in our case, first and foremost the common conditions should be ruled out and a high degree of clinical suspicion should be kept for this disease as it is often missed. Treatment of the disease should involve holistic approach including dermatologist, ophthalmologist, otolaryngologist and physician.

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