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CASE REPORT

Case Report: A case of neurogenic bladder in the setting of Behçet's disease after an initial diagnosis of multiple sclerosis [version 1; peer review: 3 approved with reservations]

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Abstract

Behçet's disease (BD) is an autoimmune vasculitis with an unclear etiology presenting with a classic triad of symptoms including oral and genital ulcers as well as iridocyclitis. A subset of BD patients exhibit neurological symptoms including psychiatric disturbances, balance problems, and voiding dysfunction, and the symptoms of BD can mimic other neurological diseases, including multiple sclerosis (MS). Differentiating between potential diagnoses is challenging due to the lack of specific tests for these disorders and the overlap between clinical symptoms and radiological findings. We describe the case of a 52 year old woman initially diagnosed with and treated for MS. From the urologic standpoint, she was treated for neurogenic detrusor overactivity with detrusor-sphincter-dyssynergia utilizing ileocecal augmentation cystoplasty with a continent stoma for intermittent catheterization. The patient was later diagnosed with BD in light of additional clinical findings.

Keywords

Neurogenic bladder, Voiding dysfunction, Micturitional disturbance, Bladder dysfunction, Detrusor-sphincter-dyssynergia, Multiple sclerosis, Behçet's disease, Multiple sclerosis

Open Peer Review

Approval Status ? ? ?

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10 Apr 2015	view	view	view
1. John T. Stoffel , University of Michigan, Ann Arbor, USA			
2. Fereydoun Davatchi , Tehran University of Medical Sciences, Tehran, Iran			
3. Bertil Blok , Erasmus Medical Center, CN Rotterdam, The Netherlands			

Any reports and responses or comments on the article can be found at the end of the article.

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Background

Behçet's disease (BD) was first described by the Turkish dermatologist Hulusi Behçet in 1937 as a triad of clinical symptoms consisting of recurrent uveitis and aphthous ulcers of the mouth and genitalia¹. BD is now known to be a multi-system autoimmune inflammatory vasculitis with an unclear etiology and a strong link to the HLA-B51 haplotype². Patients with Behçet's disease may present with a variety of symptoms, most commonly with mucocutaneous and ocular involvement thought to be vasculitic sequelae. In addition, BD may also involve the joints as well as the gastrointestinal and nervous systems³.

Approximately 5–15% of patients with BD demonstrate neurological symptoms including dementia, psychiatric disturbances, cranial nerve palsy, cerebellar ataxia and pyramidal tract signs⁴. Micturitional disturbances have been found in 5–67% of patients with neurologic BD and these have been linked to pontine lesions affecting the pontine micturition center (PMC), pontine urine storage center (PUSC), and the nucleus reticularis pontis oralis (PoO)^{1,3,5}. Generally, nervous system lesions in neurologic BD have been observed primarily in the brain stem and spinal cord¹. Post-mortem examination has demonstrated degeneration of the pyramidal tracts as well as focal necrosis of the upper tegmentum pontis, subthalamic area, and internal nucleus of the basal ganglia^{6,7}. Generally, neurological BD is progressive in 85% of patients with remission and relapse occurring in the remaining 15%⁸.

Given that the etiology of BD is incompletely elucidated, its diagnosis remains an inexact science and relies on magnetic resonance imaging (MRI), analysis of evoked potentials and cerebrospinal fluid (CSF), and clinical symptoms. CSF analysis typically yields pleocytosis with polymorphonuclear predominance in the absence of oligoclonal bands¹. Clinical presentation is usually with an acute or subacute attack followed by remission with or without sequelae, or with progression after an initial attack. Clinically, patients can exhibit a variety of symptoms, many of which overlap with other neurological and autoimmune conditions. The spectrum of symptoms includes, but is not limited to, arthralgia and arthritis, gastrointestinal tract inflammation, deep venous thrombosis, superficial thrombophlebitis, inflammation within the cardiovascular system, inflammatory problems in chest and lungs, auditory disturbances, balance difficulties, fatigue, and psychiatric disturbances^{9,10}. As a result of the lack of specificity of the above findings, serologic testing to narrow the spectrum of differential diagnoses is typically undertaken, but there is currently no sensitive or specific serologic test for diagnosis of BD itself.

Multiple sclerosis (MS), like BD, is regarded as an autoimmune disorder involving the central nervous system (CNS) and resulting in axonal demyelination. Symptoms of MS are variable in spectrum and presentation and can involve almost any aspect of the nervous system. As a result, clinical data alone may be insufficient to make the diagnosis, and supporting data come from neuroimaging, CSF analysis, and evoked potentials. MRI findings can demonstrate plaques, which, in contrast to those found in BD, can be located in white matter of the optic nerve, the basal ganglia, and close to the ventricles of the cerebellum in addition to in the brain stem and

spinal cord¹¹. CSF analysis may show pleocytosis, but in contrast with the polymorphonuclear nature of that seen in BD, pleocytosis in MS is typically lymphocytic. In addition, oligoclonal bands of IgG may be seen on electrophoresis of CSF in 75–85% of patients with MS¹². Analysis of evoked potentials demonstrates decreased amplitudes secondary to demyelination¹³. In contrast to BD, which can either present with an initial attack with or without sequelae or in a secondary progressive manner, MS is always progressive, and can follow either a relapsing remitting, relapsing progressive, or primary or secondary progressive pattern.

We describe the case of a 52 year old female who was initially diagnosed with relapsing remitting MS. This diagnosis was amended to Behçet's disease after further workup 10 years later. In the interim, the patient was diagnosed with a neurogenic detrusor overactivity and detrusor-sphincter-dyssynergia and was treated initially with anticholinergic medications, intermittent catheterization, and subsequently with ileocecal augmentation cystoplasty.

Case presentation

The patient is a 52 year old Hispanic female with a history of obsessive compulsive disorder and anorexia since adolescence who developed depression and insomnia while in her forties. In addition, she exhibited progressively worsening bilateral hand tremors, worsening dexterity, anorexia, and balance difficulties. A single-photon emission computed tomography (SPECT) scan of the brain on initial presentation in 1998 demonstrated hypoperfusion of the basal ganglia and frontal lobes. The patient was treated with steroids at the time, resulting in transient improvement of her insomnia. A MRI of the brain performed in June of 2000 demonstrated white matter lesions within the subcortical regions including the corpus callosum and periventricular areas, as well as posterior fossa atrophy. Visual and somatosensory evoked responses, as well as auditory brainstem responses, were normal. The patient declined a lumbar puncture, and thus cerebrospinal fluid was not available for analysis. Immune panels, anti-nuclear antibody (ANA), anti-neutrophil cytoplasmic antibody (ANCA), angiotensin converting enzyme (ACE), and chest X-ray were all within normal limits. Per report from the patient, an echocardiogram performed in Mexico in 2000 demonstrated minimal mitral valve prolapse, but neither the echocardiogram report nor images were available for review. At this time, her physical exam demonstrated myokymia in a cranial nerve VII distribution, right upper extremity pyramidal drift, postural and action tremors in bilateral upper extremities, and balance difficulties when asked to stand on one foot. In light of these findings, which met the Barkhoff and Tintoré criteria for diagnosis of MS, and despite the presence of a significant psychiatric component, a diagnosis of relapsing-remitting MS was made^{14,15}. Notably, the literature supports a diagnosis of MS in the setting of psychiatric disease¹⁶.

As a result of her neurological symptoms and findings on imaging, the patient was started on glatiramer acetate and intermittently remained on this medication until 2002. She was not treated with interferons given their potential to worsen depression¹⁷. Initially, her symptoms improved markedly with near-resolution of her hand tremor. However, she continued to complain of balance

difficulties and memory problems. In addition, in October 2000, she experienced vertigo and confusional episodes as well as worsening depressive symptoms, all of which responded to pulsed intravenous sumamedrol. In January 2002, she demonstrated worsening right hand dexterity with no changes in memory and operational judgment when compared with her prior visit. Repeat MRI at that time demonstrated decreased burden of disease per report. She underwent neuropsychological evaluation at this time as well, which demonstrated organic memory deficits with intact intellect. In March 2002, the patient became mentally disorganized and was admitted to the hospital for treatment and observation. At this time, her glatiramer acetate was stopped and the diagnosis of MS questioned. Repeat MRI in June 2002 demonstrated no increase in disease burden. On physical examination at this time, she continued to exhibit minimal tremor in bilateral upper extremities but no longer had pyramidal drift in her right upper extremity. As a result of her symptomatic improvement, glatiramer acetate was not restarted.

In October 2003, a MRI of the brain evidenced an increase in disease burden and symptomatically the patient exhibited worsening memory deficits. At this time, however, the patient was complaining of urgency and urge incontinence and was evaluated by other urologists. She underwent a Marshall-Marchetti-Krantz bladder neck suspension in late 2003 and subsequently developed immediate urinary retention requiring placement of a Foley catheter. As a result, she was referred to our practice for evaluation and further management. Video urodynamic evaluation in January 2004 demonstrated

neurogenic detrusor overactivity with some loss of compliance, detrusor-sphincter dyssynergia, elevated voiding pressures, poor flow, and a post-void residual of 350ml. Her first sensation occurred at 90ml bladder volume with first urge to void at 270ml and a functional bladder capacity of 340ml (Figure 1). She did not leak with cough or Valsalva maneuver. Fluoroscopy and electromyogram (EMG) demonstrated mid-urethral obstruction at the sphincter and no vesicoureteral reflux.

The patient desired to be free of urethral catheter drainage of her bladder, which was inconvenient and difficult given her limited manual dexterity. As a result, the patient underwent ileocecal augmentation cystoplasty with continent stoma in February 2004 without complications¹⁸. Her post-operative course was unremarkable and on subsequent office visits, she was doing well with much improved bladder symptoms consisting primarily of infrequent bladder spasms and occasional urinary tract infections with no urethral incontinence. These were successfully treated with intravesical anticholinergic medication and antibiotics. On subsequent video urodynamic evaluation performed in June 2010 an augmented bladder with normal compliance and 500ml capacity was demonstrated. There was no evidence of uninhibited contractions, urgency, vesicoureteral reflux, or leakage with cough or Valsalva.

In addition to the above symptoms, the patient was noted to be complaining of intermittent gastric upset, joint pain and swelling, muscle aches, oral, vaginal, and rectal ulcers, and intermittent

Figure 1. Initial urodynamic tracing of the presented patient. “EMG” – electromyogram “Pves” – intravesical pressure, measured in centimeters of water; “Pabd” – intra-abdominal pressure, measured in centimeters of water; “Pdet” – detrusor pressure, measured in centimeters of water; “Flow” – urinary flow, measured in milliliters per second. The arrow corresponds to the functional bladder capacity of 340 milliliters.

fevers over the past several years. As a result, she was referred for rheumatologic workup in early 2010 which was remarkable for an elevated erythrocyte sedimentation rate (ESR) and C-reactive protein (CRP), but with all other tests, including ANA, anti-cardiolipin antibody, rheumatoid factor, ACE, HLA-B27, antithrombin III, and complement studies within normal limits. Given the symptoms that she was exhibiting in addition to the neurological symptoms she initially presented with, the patient was diagnosed with fibromyalgia and Behçet's disease and started on azathioprine and vitamin D supplementation, on which she remains to this day with stable symptoms.

Discussion of treatment and management

The above patient represents a diagnostic dilemma resulting in a change in diagnosis during the course of her treatment. Initially, the patient was diagnosed with MS on the basis of imaging findings as well as clinical symptoms that were consistent with the diagnosis, including CNS plaques in a distribution typically associated with MS, cranial nerve palsy, balance difficulties, and upper extremity tremors. However, CSF could not be obtained for evaluation, limiting the ability to differentiate between MS and other neurologic disorders. Furthermore, the patient initially presented with psychiatric symptoms that are atypical in the setting of MS, though an association between MS and psychiatric symptoms has been described¹⁶. Finally, additional serologies demonstrated no evidence for autoimmune disease or other findings that would suggest an alternate diagnosis. In addition, evaluation of evoked potentials was normal, which is unusual in the setting of MS, as 85% of MS patients have abnormal visual evoked potentials, 77% have abnormal somatosensory evoked potentials, and 67% have abnormal brainstem auditory evoked potentials. Nevertheless, it was not unreasonable to make an initial diagnosis of MS given the clinical and laboratory evidence at the time.

During the course of the patient's treatment, additional symptoms became manifest that resulted in a change in diagnosis from MS to BD. These symptoms included oral and anogenital ulcers, gastrointestinal upset, and joint pain and swelling. Serologies continued to be unremarkable save for a transient increase in erythrocyte sedimentation rate and CRP. Together with the previously described symptoms and findings on imaging, this led to a change in diagnosis to neurologic BD. Lamentably, no evaluation of HLA-B51 haplotype was undertaken in this patient, as the Bw51 allele is closely associated with BD¹⁹. In addition, the absence of CSF for analysis further hindered an early, accurate diagnosis in this patient.

The patient's initial treatment, based on the MS diagnosis, included pulsed steroids and glatiramer acetate. It is likely that the patient derived benefit from steroids, as these were used over short periods during symptom exacerbations and have demonstrated efficacy in treatment of both MS and BD symptoms^{20,21}. In contrast, it is less clear whether the patient derived significant benefit from the glatiramer acetate, as there appear to be no studies evaluating its use in BD, and while its use in MS is established, the relapsing/remitting nature of the patient's symptoms and the possibility of a wrong diagnosis calls into question the efficacy of the drug as administered to this patient.

From a urologic standpoint, the patient was not properly diagnosed by her urologists. She carried a clear neurologic diagnosis and her complaints were "overactive" in nature, not centered on stress urinary

incontinence. The retropubic suspension aggravated her retention until she sought further medical care. She presented with neurogenic detrusor overactivity and detrusor-sphincter-dyssynergia along with symptoms that may be found in patients with both MS as well as BD²². Few studies exist examining micturitional disturbances in BD patients and the incidence of micturitional disturbances in patients with BD ranges between 5–67%^{23–25}. In one study comprising eight patients with BD evaluated for lower urinary tract symptoms, seven demonstrated bladder dysfunction. The most common urodynamic finding was detrusor overactivity demonstrated in six patients, which is consistent with our findings. Bladder biopsies from these patients demonstrated blood vessel wall thickening and inflammatory infiltration of the lamina propria, consistent with prior findings by the same authors^{26,27}. Unfortunately, no tissue samples from our patient were available for pathologic analysis, preventing a comparison with the findings in the literature. This reported pathologic finding could result in loss of detrusor compliance.

A larger study evaluating 24 subjects with neurological BD demonstrated a 50% incidence of detrusor overactivity and 12.5% incidence of detrusor-sphincter-dyssynergia³. These patients generally had high post-void residuals, early first sensation, and low-normal bladder capacities, which mimic the findings in our patient. In addition, the authors mapped the presence of CNS lesions in their subjects, demonstrating the majority in the brain stem with a subset in the cerebrum, which is consistent with other studies¹. Our review of the images revealed no lesions in areas typically affected by BD or in areas that may affect micturition as described above. The high incidence of overactive bladder symptoms demonstrated in our patient is also echoed in several other studies^{5,23,24,28}, as are our findings on urodynamics¹.

Our patient initially received anticholinergic medications for her overactive bladder symptoms with incomplete relief. Recommendations in the literature suggest an initial trial of anticholinergic therapy with the goals of preventing uninhibited contractions and lowering intravesical pressure, parameters which have been shown to improve with anticholinergic treatment in up to 75% of patients^{3,26}. Our patient developed significant dry skin with oral anticholinergic medication and asked to be taken off the medication. While intravesical instillation of anticholinergic medications is an option that has been shown to be effective in patients with bladder symptoms due to neurological BD, this was not attempted in our patient²⁴. Given the incomplete response to oral medications in our patient, as well as the desire to be free of an indwelling catheter in the setting of limited dexterity, ileocecal augmentation cystoplasty with a continent stoma was performed with a notable decrease in urgency symptoms and increase in bladder capacity postoperatively. Several studies have discussed the use of augmentation cystoplasty in patients with neurological BD resulting in improvements in bladder symptoms, continence status and renal function^{3,23,26}. In agreement with these studies, we found our patient to have no uninhibited contractions as well as improved bladder compliance and increased bladder capacity after augmentation cystoplasty.

Conclusions

In summary, we describe a case of BD initially diagnosed as MS in a female with a neurogenic bladder secondary to her neurologic disease. The treatment of her neurologic disease was in line with

the accepted standard of care for her diagnoses at the time they were made, as was the treatment of her neurogenic bladder. This case demonstrates the current challenges to accurate diagnosis of certain neurologic conditions and highlights the need for development of further sensitive and specific assays to facilitate these diagnoses, along with prudent use of urodynamic testing. Furthermore, this case demonstrates the efficacy of bladder augmentation with a continent catheterizable stoma in the setting of neurogenic detrusor overactivity in a patient with neurologic BD.

Consent

Written informed consent for publication of their clinical details and/or clinical images was obtained from the patient.

Author contributions

AWP reviewed all patient information, performed a literature review, participated in intellectual discussion pertinent to the conception

of the report, and drafted the manuscript. TBB is the physician of the patient presented herein, provided all patient information, and participated in intellectual discussion pertinent to the conception of the report. Both authors have agreed to the final content of the manuscript.

Competing interests

No competing interests were disclosed.

Grant information

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I confirm that the funders had no role in study design, data collection and analysis, decision to publish, or preparation of the manuscript.

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Bertil Blok

Department of Urology, Erasmus Medical Center, CN Rotterdam, 3015, The Netherlands

The manuscript 'Case Report: A case of neurogenic bladder in the setting of Behçet's disease after an initial diagnosis of multiple sclerosis' is an interesting paper. However, I have some comments addressed below.

The case report is a readable report on an interesting topic. However, the authors do not report on any vaginal child delivery nor do they mention the BMI of the patient. Both are risk factors for stress urinary incontinence. It is very possible that before the MMK a mixed urinary incontinence was present and in retrospect it is always easy to say that the previous physicians did not do a good job.

The blaming distracts from the main important message that patients with a neurogenic bladder are different from patients without a neurogenic bladder. Both referring physicians and physicians who provided the irreversible surgical treatment were responsible for the patient. This means that also the general practitioner and neurologist should be informed and know to whom they send their patients to. On a regular basis we observe maltreatment because the referring physician did not care to refer his or her patient specifically to an expert in the field.

Some attention should be given to treatment with botulinum toxin and midurethral tapes, which were also around when the bladder augmentation was given.

Is the background of the case's history and progression described in sufficient detail?

Yes

Are enough details provided of any physical examination and diagnostic tests, treatment given and outcomes?

Yes

Is sufficient discussion included of the importance of the findings and their relevance to

future understanding of disease processes, diagnosis or treatment?

Yes

Is the case presented with sufficient detail to be useful for other practitioners?

Yes

Competing Interests: No competing interests were disclosed.

Reviewer Expertise: urinary incontinence, neurourology

I confirm that I have read this submission and believe that I have an appropriate level of expertise to confirm that it is of an acceptable scientific standard, however I have significant reservations, as outlined above.

Reviewer Report 05 June 2017

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Fereydoun Davatchi

Rheumatology Research Center, Department of Internal Medicine, School of Medicine, Tehran University of Medical Sciences, Tehran, Iran

First, the authors have to prove that this patient has Behcet's Disease. For that, they have give the exact medical history of the patient. We know approximately when the patient started to have Neuro-psychiatric manifestations, but we don't know when the oral ulcers started and how long after the genital ulcers appeared.

Second, we have to know how long each attack of oral ulcer took to disappear. Then we have to know how long the duration between the two attacks was. Then after, we have to know how many ulcers appeared in each attack. Finally we have to know the exact clinical manifestations of the ulcers and their progression until their disappearance.

The same has to be given for genital ulcers.

It is primordial to remember that not any oral or genital ulcer is an aphthous ulcer, and only an aphthous ulcer can be used as a diagnostic criterion. There are many oral or genital ulcers that may resemble an aphthous lesion, to the eyes of a non-expert. It is why for case reports like this, a high definition picture of the lesion is essential to be sure of the nature of the lesion.

Once it is accepted that the oral lesion is an aphthous lesion, the authors have to prove that the genital ulcers were also aphthous ulcers.

Once the presence of oral and genital aphthous ulcers is proved, one can say that the patient may

have a Behcet's Disease, because the patient fulfills the International Criteria for Behcet's Disease (the ICBD). However, as said before, the patient may not have Behcet's Disease. To be sure, one has to not find any other reason for the presence of the symptoms together.

When it is sure that the patient has Behcet's Disease, one has to show that the neurological manifestations are related to Behcet's Disease. A patient can have Behcet's Disease and another neurological disease like Multiple Sclerosis. In this case, the patient refused an examination of the Cerebrospinal fluid (CSF).

Is the background of the case's history and progression described in sufficient detail?

No

Are enough details provided of any physical examination and diagnostic tests, treatment given and outcomes?

No

Is sufficient discussion included of the importance of the findings and their relevance to future understanding of disease processes, diagnosis or treatment?

No

Is the case presented with sufficient detail to be useful for other practitioners?

No

Competing Interests: No competing interests were disclosed.

Reviewer Expertise: Behcet's Disease

I confirm that I have read this submission and believe that I have an appropriate level of expertise to confirm that it is of an acceptable scientific standard, however I have significant reservations, as outlined above.

Reviewer Report 30 January 2017

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John T. Stoffel

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This is an interesting report on a patient with Behcet's disease and urological symptoms. The neurological description of presentation, progression, and treatment is outstanding. The case report nicely reviews Behcet's disease for the reader in a clear and concise way. The authors should be commended for highlighting this uncommon disease. The presentation of the

urological symptoms and the relationship to Behcet's disease in this patient are not as clear. My comments:

1. The introduction notes that the patient was treated with an ileocecal augment/continent stoma for neurogenic detrusor overactivity and DSD. The description, however, notes that she was in retention and could not void after a MMK procedure. Presenting fluoro images would be helpful for the reader to better understand how the diagnosis of DSD was reached versus post procedural obstruction. By the history, she could not void after the MMK making it more likely that this is contributing to her retention.
2. The discussion notes that the patient was not properly diagnosed by her urologists. Is it possible that she did have mixed incontinence prior to MMK and then developed complications from this procedure rather than a missed diagnosis of neurogenic DO? More data could be presented to highlight educational opportunities on what the authors feel the work up could have included prior to MMK to avoid the complication and to better work up neurogenic bladder patients.
3. The authors could also touch on the role of Botox in treating neurogenic DO.

Is the background of the case's history and progression described in sufficient detail?

No

Are enough details provided of any physical examination and diagnostic tests, treatment given and outcomes?

No

Is sufficient discussion included of the importance of the findings and their relevance to future understanding of disease processes, diagnosis or treatment?

No

Is the case presented with sufficient detail to be useful for other practitioners?

No

Competing Interests: No competing interests were disclosed.

I confirm that I have read this submission and believe that I have an appropriate level of expertise to confirm that it is of an acceptable scientific standard, however I have significant reservations, as outlined above.

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