Sacral insufficiency stress fracture as etiology of positional autonomic dysreflexia: Case report

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The medical literature is replete with case reports of the syndrome known as autonomic dysreflexia. Although the majority of cases are known to be induced by either bladder or bowel distention, there does exist a small number of cases in which the inciting stimulus is more obscure. In such cases, a comprehensive medical evaluation is necessary to ensure proper identification of the source of irritation and the appropriate medical management of the patient. We present a patient with a heretofore unreported suspected etiology of autonomic dysreflexia, axial loading of a sacral stress fracture.

Keywords: autonomic dysreflexia; spinal cord injury; tetraplegia; stress fracture

Case report

A 41 year old male with a traumatic C5 complete tetraplegia of 25 years duration presented to the spinal cord injury unit for his annual physical examination and evaluation of recurrent urinary tract infections (UTIs). The patient was known to have a neuropathic bladder secondary to his spinal cord injury (SCI) and had undergone three previous sphincterotomies, utilizing a condom catheter for bladder drainage. He also had a neuropathic bowel for which a digital stimulation and suppository bowel program was used every three days without difficulty.

The patient had no medical complaints on admission. His physical examination was consistent with a C5 complete tetraplegia. His laboratory results: (complete blood count (CBC), chemistry and prostate-specific antigen) were unremarkable. Following admission, a routine urodynamic study was performed which revealed a large-capacity, low pressure bladder. The urological recommendation was for placement on an intermittent catheterization program, however, the patient instead consented to the use of an indwelling Foley catheter.

On the day following the urodynamic study, during an assisted transfer from bed-to-wheelchair, the patient and nursing staff reported hearing a 'popping sound' suspected in the thoracic region of the back. The patient immediately experienced a flushing of his upper body along with diaphoresis, piloerection, hypotension (BP=87/53) and tachycardia (Pulse of 115). The episode resolved within minutes after the patient was returned to his bed and was placed in a recumbent position. A complete physical examination

did not reveal any changes from those present on admission. The patient's indwelling Foley catheter was removed and was replaced. Thoracic and lumbosacral spine radiographs were obtained and revealed severe osteopenia but no evidence of fracture or subluxation. The urine culture grew *Serratia marcescens* present in <100K colonies and the patient was started prophylactically on an oral antibiotic.

On subsequent days whilst lying supine in bed, the patient experienced no further such episodes but he did complain of shortness of breath and palpitations. He had a cardiac dysrhythmia, and a 12-lead electrocardiogram confirmed the presence of a bigeminy-trigeminy rhythm with unifocal premature ventricular contractions (PVCs), incomplete right bundle branch block and a previously noted right axis deviation. An echocardiogram was normal except for dysrhythmia. Chest radiographs revealed mild pulmonary edema. Cardiology consultants determined that this rate-controlled asymptomatic rhythm did not require medical intervention. The pulmonary edema resolved spontaneously in subsequent days.

The patient proceeded to experience autonomic dysreflexia (AD) with hypertension (occasional hypertensive episodes), tachy- or bradycardia, and cephalgia along with the previously noted symptoms when placed in a seated position. His symptoms resolved upon reclining to the supine position.

Further examination revealed a mildly enlarged right testis and ultrasonography demonstrated epididymal cysts and a small hydrocele. Follow-up urine cultures grew *Serratia* and the patient was treated for 21 days with appropriate intravenous antibiotics. Despite antibiotic treatment and subsequent negative urine cultures, the patient continued to experience episodes of positionally-induced AD.

During the course of evaluation and treatment of the patient's AD, multiple etiologies and treatments were pursued, including: bladder and bowel decompression, inspection of the skin for breakdown or irritation, assessment of the position of the scrotum while sitting, examination for intra-abdominal pathology (including KUB and ultrasonography for gastrointestinal, kidney or gallbladder disease) and laboratory evaluation (CBC and serum chemistries). These evaluations revealed no evidence of pathology. Of note on physical examination was the absence of AD symptoms on passive flexion of the hips while the patient was supine in bed but present when the patient was seated. Laboratory studies revealed an elevated alkaline phosphatase level and therefore pelvic and hip radiographs were obtained to evaluate the possible presence of heterotopic ossification (HO). These radiographs demonstrated an old fracture at the right anterior superior iliac spine, diffuse osteopenia, osteitis pubic symphysis, and an old bone infarct of the right femur. There was no evidence of HO or of a sacral fracture (Figure 1).

A Technetium 99 m bone scan was obtained which revealed increased uptake in the cervical spine (the site of the original SCI trauma), the sacroiliac joints and

the sacrum. The sacral pattern of uptake was in an H-pattern most consistent with a stress fracture of the sacrum¹ (Figure 2). An Indium WBC-tagged study was performed to rule out osteomyelitis of the pelvis; there was no evidence of infection.

The patient was treated with a progressive sitting program consisting of a gradual increase in both the angle of incline and the time spent sitting up in his wheelchair. He was ultimately able to tolerate sitting for prolonged periods of time without symptoms of AD. He was discharged home with occasional mild episodes of AD which eventually resolved.

Discussion

Autonomic dysreflexia (AD) may be seen in approximately 85% of spinal cord injured patients with injuries above the major splanchnic outflow in response to noxious stimuli below the level of the lesion.²⁻⁷ Observation, experimental models and postulates of the pathophysiologic mechanism(s) can be found in published literature. Most suggest the mechanism involves the loss of the descending central sympathetic inhibitory activity. Although numerous stimuli such as tight clothing,⁷ ingrown toenails,⁸

Figure 1 Roentgenogram of pelvis demonstrates osteopenia without evidence of sacral fracture

Figure 2 Delayed posterior image of sacral portion of whole body Technetium 99m bone scan demonstrates increased bone uptake in H-pattern characteristic of sacral insufficiency stress fracture

pulmonary emboli,⁹ or an acute surgical abdomen^{10,11} have been cited as initiating AD, clinical observations reveal bladder and bowel distention as the two most frequent stimuli causing AD in the clinical setting.^{2,7}

Postural AD has been previously reported. Abouleish *et al*¹² described AD stimulation by movement in a parturient, however, uterine contractions could have been the inciting stimuli in this case.^{2,13} Khurana reported three subjects with orthostatic hypotension-induced AD, alerting the clinician to the potentially counterproductive effect of sitting the patient upright and to the need for medical management of hypotension-induced AD.¹⁴ Although the symptoms were effectively treated in that series of patients, the stimulus for hypotension was presumed to be autonomic insufficiency; no evaluation of bony pathology was recorded. Thus, it is uncertain as to the underlying pathology of AD in these long-term tetraplegic patients who would undoubtedly have had osteopenia. Thompson and Witham reported a patient who noted AD symptoms with catheter changes, sudden changes of position, and following the administration of enemas.³

The potentially serious medical sequelae of AD as demonstrated in this case and others, cardiac dysrhythmia⁵ and pulmonary edema,¹⁵ compels the clinician to identify and irradiate the noxious stimulus(i). After thorough evaluation, we suspected two stimuli to be the most likely cause of this patient's AD, a urinary tract infection and a possible epididymitis. The appropriate treatment of these infections along with the positional nature of the AD argued against either of these as being the etiology of his symptoms. The clinical history of a 'popping sound' in the vertebrae just prior to the first episode of AD raised the question of a fracture and/or subluxation of the vertebrae as the symptom's source. Negative radiographs of the thoracic and lumbosacral spine made this unlikely. With the elevated alkaline phosphatase level, heterotopic ossification (HO) was next suspected as the stimulating agent, yet this was not confirmed by the radiographs or the bone scan. The Technetium bone scan did reveal the characteristic H-pattern felt to be diagnostic of a stress fracture of the pelvis.¹ Once the patient's sacral stress fracture was diagnosed, the appropriate treatment regimen could then be utilized to manage his AD.

Conclusion

The increased risk for fracture of the extremities as a result of osteopenia in individuals with spinal cord injury is well-documented.¹⁶ What may not be appreciated, however, is the degree to which this increased risk for fracture may also exist in other

osseous structures. The patient presented in this case apparently sustained sufficient trauma in the recent past to cause a pelvic stress fracture even though there was no history of any such event(s). Plain radiographs failed to reveal evidence of fractures of the sacrum but a stress fracture was demonstrated by the bone scan. The necessity for continuing to search for the cause of an episode of AD in an SCI patient has been demonstrated in this case. It is only after the inciting stimulus has been identified that the rendering of appropriate medical treatment can be assured.

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