

Case Reports

Bilateral Gluteal Dyskinesia: Discussion of a Rare Movement Disorder

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Abstract

Background: Involuntary movements of gluteal muscles have rarely been reported.

Case Report: This 46-year-old female with pelvic endometriosis developed involuntary rhythmic movements in the left gluteus maximus, which within a year became bilateral. The movements gradually increased in intensity and interfered with ambulation. Electromyography, at rest, demonstrated almost continuous periodic gluteal discharges, with left-sided discharges seeming to lead to those on the right. OnabotulinumtoxinA injections into the gluteal muscles improved the movements.

Discussion: A rare and previously unreported form of gluteal involuntary movements with periodic electromyographic discharges is described. The cause is uncertain. The differential diagnosis of this unusual movement disorder is discussed, with the most likely diagnosis being myoclonus.

Keywords: Painful legs moving toes, painless legs moving toes, endometriosis, botulinum toxin, botulinum neurotoxin, onabotulinumtoxinA

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Introduction

Focal symptoms and signs in the gluteal region can arise in a variety of settings, including iatrogenic nerve damage secondary to intragluteal injections,¹ hip arthroplasty,^{2,3} infiltration with various neoplastic lesions,⁴ and piriformis entrapment syndrome.⁵ Involuntary movements localized to the gluteal muscles have rarely been reported in the literature, and include post-traumatic focal dystonia with axial dystonia,⁶ focal dyskinesia of the gluteal, low back, and abdominal muscles likely due to abdominal surgery,⁷ and bilateral widespread twitching of the posterior thighs and calves, including the gluteal regions, believed to represent Isaac's syndrome.⁸ We describe a previously unreported involuntary movement confined to the gluteus maximus (GM) muscles.

Case Report

The patient, a 46-year-old female, had complained of increasing difficulty in walking over the past year. Her husband described involuntary movements of her “buttocks”, which began about 10 years

previously without apparent trigger. The movements started first on the left side, but within a year also involved the right side. The movements gradually worsened and particularly bothered her at night and during ambulation. Her past history was significant for pelvic endometriosis, which was detected on magnetic resonance imaging (MRI) performed 3 years prior to our evaluation. It showed extensive bilateral adnexal involvement, suggesting long-standing disease. The patient underwent hysterectomy and bilateral salpingo-oophorectomy 1 year later. The character of her involuntary movements did not change following surgery.

Her other medical problems included sleep apnea, obesity, gastric bypass surgery, and pelvic surgery 3 years previously for extensive endometriosis. The patient denied a history of trauma to the lower back or thighs, and had no leg or lower back pain.

On examination, her vital signs were normal. The general medical examination disclosed no abnormalities. Her neurological examination was normal, with the exception of continuous involuntary rhythmic and semi-rhythmic movements of the GM muscles on both sides. The movements were present at rest but fluctuated in intensity (Video 1).



Video 1. Bilateral gluteal dyskinesia and botulinum toxin injection. Bilateral rhythmic gluteal movements associated with periodic discharges in electromyography and electromyography-guided onabotulinumtoxinA injection.

The lower part of GM muscles was predominantly involved. Movements increased in intensity during ambulation, but did not seem to spread to other muscles. The movements were painless. There was no associated muscle weakness.

Electromyography (EMG) of the GM muscles, at rest, showed regular periodic and semi-periodic burst discharges at a frequency of 2–5 Hz. The burst duration varied from 50 to 100 ms. No denervation potentials were noted. However, the EMG study was limited due to the patient's significant discomfort. On dual-channel EMG recording, the left-sided discharges often preceded the right-sided discharges by less than 100 ms (Figure 1). EMG of the gluteus medius, long head of the biceps femoris, and lumbar paraspinal muscles showed no denervation

potentials and a normal recruitment and interference pattern. Nerve conduction study of the left tibial nerve to the abductor hallucis was normal. Left tibial F-waves were also normal. An MRI of the thoracic and lumbar spine showed mild degenerative disc disease, most pronounced at L5–S1. Computed tomography (CT) of the chest showed non-specific ground-glass opacity in the lower lobe of the left lung. Pelvic MRI showed resolution of the bilateral adnexal hemorrhages seen on the MRI scan 3 years previously (before surgery). A focused MRI of both thighs covering the gluteus muscles showed no abnormality. A positron emission tomography (PET) scan was performed, which was normal.

Patient received injections of onabotulinumtoxinA into the GM muscles under EMG guidance. Injections were performed through a special EMG/injection hollow needle at the point of maximum discharge. These points were located in the more distal part of the GM muscle. The dose was 180 units/side, given at three sites of the areas of maximum movement. After 1 week, the patient reported marked reduction of her gluteal movements and steadier ambulation. After 6 months, the patient returned to the clinic because of increased intensity of the movements. The movements were again confined to GM muscles. A repeat injection with the same dose improved the movements significantly. The patient did not wish to have a post-injection EMG study. A repeat chest CT at 6 months showed no change in the left lung lesion, which was considered benign and non-specific.

Discussion

This patient presented with unique rhythmic and semi-rhythmic movements in both glutei, which correlated with bursts of grouped

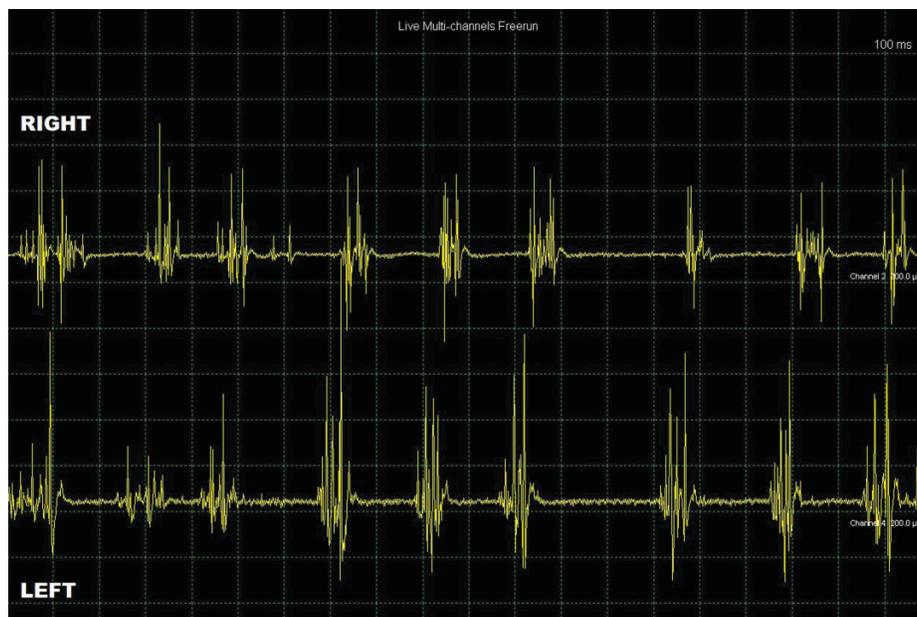


Figure 1. Dual-channel Electromyography. The electromyograph shows bilateral periodic discharges within the gluteal muscles, with the left side often preceding the right side by less than 100 ms.

repetitive, action potentials with fluctuating amplitude and frequency in EMG. The movements were confined to the GM muscle, and EMG and nerve conduction studies showed no evidence of any other muscle involvement, especially those innervated by the sciatic and superior gluteal nerves.

The differential semiology of our patient's movements includes myoclonus, tremor, chorea, dystonia, and some other rare semi-rhythmic movements. Myoclonus can be central or peripheral (less common). Spinal myoclonus is characteristically rhythmic. Myoclonus will be discussed further below. Tremor by definition is a rhythmic movement disorder. The character and distribution of movements, however, do not match distribution of any known tremors. Enhanced physiologic tremor and drug-induced tremor are two common forms of tremors. Both have faster frequency and predominantly distal distribution. Parkinson's disease tremor and essential tremor also usually affect the hands and forearms, and have a different character. Chorea is a common movement disorder and is often drug induced. Drug-induced chorea frequently affects the facial musculature. The movements have a flow, wax and wane in frequency and intensity, and are more distal than proximal. In neurodegenerative disorders such as Huntington's disease patients often manifest other symptoms such as dystonia, cerebellar and gait disorder, and cognitive decline.

Dystonic movements are twisting and turning; with co-contraction of agonist and antagonist muscles. Clinical and EMG rhythmicity such as seen in this case is highly unusual.

The cause of our patient's gluteal movements remains elusive. Limitation of movement to GM muscle suggests isolated dysfunction of the inferior gluteal nerve. Colorectal carcinoma is always a concern with symptoms related to isolated inferior gluteal nerve dysfunction.⁴ Our patient's work up, including a total body PET scan and the aforementioned MRIs, did not disclose any colorectal pathology. Association with extensive pelvic endometriosis is intriguing, but the cause and effect cannot be established since pelvic surgery did not alter the movements. Endometriosis is known to affect tissues out of the pelvis and abdominal cavity.⁹ Her stationary lung lesion is a suspect for this pathology. In neural tissue, isolated involvement of sciatic nerve has been described before with deep endometriosis.¹⁰

The patient's gluteal movements probably represent a form of myoclonus. Spinal myoclonus is often rhythmic. A central generator in the lower cord could have caused the patient's time-locked bilateral

movements. The etiology remains elusive since her neuroimaging studies showed no abnormality. Alternatively, although highly unusual, constant irritation of one peripheral nerve could cause a central loop (spinal); this can generate time-locked movements in the contralateral muscle. Contralateral development of movements have been reported for rare unilateral movements with periodic or semi-periodic EMG discharges and attributed to development of a central loop.¹¹

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