

**LETTER TO THE EDITOR****To THE EDITOR****Recurrent Migrating Focal Myositis: A Pediatric Case Report**

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Focal myositis (FM) is a localized inflammatory pseudotumor characterized by sudden-onset inflammation in one skeletal muscle.<sup>1,2</sup> Contrary to other forms of inflammatory myositis, FM is usually not associated with systemic symptoms or muscle weakness, though fever is occasionally reported.<sup>1</sup> FM is typically isolated to one muscle and resolves spontaneously within weeks without lasting symptoms. Cases of relapsing symptoms have been reported in up to 41% of adult patients,<sup>3</sup> but relapsing FM remains poorly described in children. Here, we present the first case, to our knowledge, of bilateral migrating relapsing FM in a child.

A 12-year-old male, born in Canada to a Rwandan mother and Haitian father, presented with gradual bilateral neck pain and neck enlargement over the course of 4–10 days. The patient showed no systemic symptoms or muscle weakness. Pregnancy and delivery were uneventful. Psychomotor development was normal. The patient reported three episodes of myalgia in the preceding year – the first in the left thigh, the second in the right foot, and the third in both thighs. The patient consulted in person for the first two episodes; on both occasions, inflammatory markers were slightly elevated, creatinine kinase levels were normal, and magnetic resonance imaging (MRIs) of the affected muscles showed T2 hyperintensity with limited involvement of surrounding connective tissue, consistent with myositis.<sup>3,4</sup> All episodes resolved within 2 weeks. The patient was otherwise healthy and took no medications at home. Family history was unremarkable.

The patient, who weighed 54.4 kg, was started on Naproxen 500 mg twice daily for 3 weeks. Like his previous episodes, the patient had mildly elevated inflammatory parameters but normal creatinine kinase and aldolase levels. MRI of the sternocleidomastoid muscle (SCM) muscles showed T2 hyperintensity, consistent with his prior visits. His symptoms improved throughout his 6-day hospitalization. Extensive auto-immune and infectious workups (including a comprehensive myositis-specific autoantibodies panel) were negative. Because of the risks and potential morbidity involved in a SCM biopsy, no biopsy was conducted. Given the patient's rapid clinical improvement, lack of systemic symptoms, and normal ancillary investigations across multiple visits, alternative diagnoses such as vasculitis and inflammatory myositis were excluded. On follow-up, the patient remained asymptomatic 2 weeks after the end of his Naproxen regimen. A detailed clinical and paraclinical description of the patient's

myalgia episodes is provided in Table 1. Figure 1 presents MRI images of the muscle groups involved in all three hospital visits.

FM is a rare disease that usually presents as self-resolving inflammation of a single muscle, typically the calf, forearm, or thigh. Atypical presentations have been reported, although SCM involvement has only been noted in adults so far.<sup>1,2,5</sup> Systemic symptoms and muscle weakness are usually absent. The disease affects both sexes equally and is most common in middle-aged adults, though individuals of all ages can be affected.<sup>1</sup>

The disease has no known etiology. Nerve lesions, infectious diseases, neoplasms, and immune disorders – notably Behcet's disease – are proposed triggers for FM.<sup>1</sup> Some reported cases of FM evolve into polymyositis (PM) or dermatomyositis (DM), but such an evolution is rare and may represent a misdiagnosis of an early form of PM that manifests as localized nodular myositis.<sup>2,4</sup> The relationship between FM and other inflammatory pseudotumors, such as idiopathic orbital inflammation, has not been studied. Whether these entities represent separate conditions or different manifestations of a common underlying etiology remains unknown.

The disease course is variable, with relapse occurring in up to 41% of adult cases.<sup>3</sup> Bilateral relapses have only been reported in adults.<sup>7</sup> Clinical evolution is benign; however, a diagnosis of FM may signal an underlying disease. In a 2018 study, FM was associated with immune-mediated inflammatory diseases (IMIDs) in up to 32% of patients, as well as with neoplasia in 24% of patients and radiculopathy in 11% of patients.<sup>3</sup> Prompt diagnosis of FM may thus help discover treatable underlying conditions.

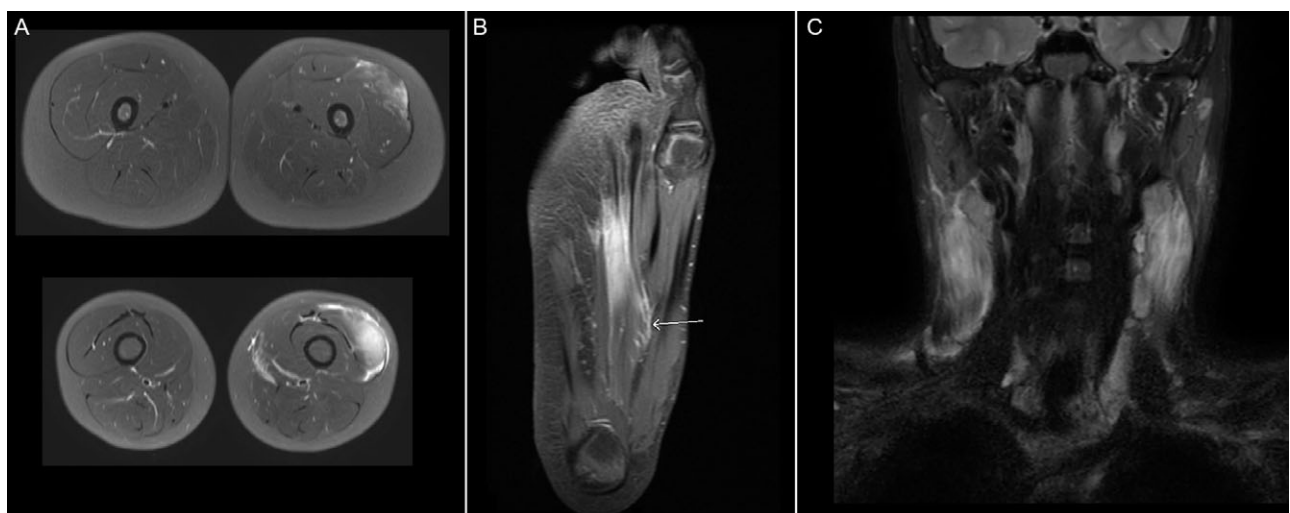
FM is a diagnosis of exclusion. Efforts have been made to build a diagnostic score, but no consensus has been reached yet on definite criteria.<sup>3</sup> Diagnosis relies on a holistic evaluation of clinical presentation, imagery, laboratory results, and pathology. Biopsy can help rule out certain alternative diagnoses; however, since histological analysis is generally non-specific and occasionally presents atypical features, some authors suggest that biopsies only be performed in the presence of systemic symptoms or persistent inflammation despite appropriate treatment.<sup>2,6</sup>

FM remains poorly studied in children. We report an unusual case, where a 12-year-old child presented with bilateral relapsing migrating FM. His symptoms and negative ancillary investigations were consistent with cases of pediatric FM.<sup>1,5</sup> Moreover, his history of localized episodes with rare systemic symptoms, no muscle weakness, and persistently normal creatine kinase levels would be highly unusual for PM/DM, vasculitis, or a genetic condition such as PAPA or SAPHO. MRI results further supported our diagnosis. Although a biopsy was not performed, the non-specific histological findings of FM would not have provided much diagnostic value in this patient's case, given the lack of systemic symptoms on multiple visits.<sup>2</sup>

**Table 1: Detailed clinical and paraclinical description of the patient’s myalgia episodes**

	1st episode 2019	2nd episode 2019	3rd episode 2020	4th episode 2020
Clinical findings	Gradual onset of pain and swelling of left thigh over 8 days Limping Non-sustained fever of 39°C on admission Resolution within days	Sudden onset of pain, swelling, and erythema on the plantar face of the right foot, 1 day after finishing ATB regimen for 1st episode No fever Spontaneous resolution within a day	Pain in both thighs Patient consulted via telemedicine Fever unknown Spontaneous resolution within 2 weeks	Gradual onset of pain and swelling of neck bilaterally over 4–10 days No weakness No fever Resolution within 2 weeks
Laboratory results	Highest CK 117 U/L (normal < 250 U/L) Highest CRP 60 mg/L (normal < 2 mg/L) Highest ESR 35 mm/h (normal < 10 mm/h) No eosinophilia	Highest CK 111 U/L (normal < 250 U/L) Highest CRP 3.1 mg/L (normal < 2 mg/L) Highest ESR 26 mm/h (normal < 10 mm/h) No eosinophilia ANA, ENA, antiphospholipid Ab, TSH, ferritin, RF, C3/C4, immunoglobulins, EBV IgM, CMV IgM all normal/negative	–	Highest CK 117 U/L (normal < 250 U/L) Highest CRP 21.6 mg/L (normal < 2 mg/L) Highest ESR 30 mm/h (normal < 10 mm/h) No eosinophilia ANA, ENA, anti-DNA, Hb electrophoresis, aldolase, toxoplasmosis Ab, cysticercosis Ab, schistosomiasis Ab, strongyloides Ab, trichinosis Ab, toxocara Ab, EBV IgM, CMV IgM, extensive myositis-specific autoantibodies panel all normal/negative
Imaging results	Thigh MRI: many T2 hyperintensities in left quadriceps muscle; small quantity of liquid around fascias; suspected punctiform focus in left adductor magnus	Right foot MRI: T2 hyperintensity of flexor digitorum brevis muscle toward myotendinous junction; T2 hyperintensity of adjacent lumbrical muscles; T2 hyperintensity of plantar aponeurosis	–	Neck U/S: enlargement and hypoechogenicity of both SCM, diffuse and not well defined Neck MRI: diffuse enlargement and T2 hyperintensity of both SCM, with diffuse enhancement and reactional lymph nodes Thigh MRI (in search of residual myositis for prospective thigh biopsy): no identified myositis focus
Treatment	Four days of IV cefazolin and IV clindamycin, followed by 8 days of oral cephalixin for suspected pyomyositis	No treatment	Cetirizine for suspected urticaria, no reported effect	Three weeks of oral Naproxen

Ab = antibody; ANA = antinuclear antibodies; ATB = antibiotic; CK = creatine kinase; CMV = cytomegalovirus; CRP = C-reactive protein; EBV = Epstein–Barr virus; ENA = extractable nuclear antigen; ESR = erythrocyte sedimentation rate; Hb = hemoglobin; MRI = magnetic resonance imaging; SCM = sternocleidomastoid muscle; TSH = thyroid stimulating hormone.



**Figure 1:** MRI images of muscle groups involved during respective episodes of myalgia. (A) Axial T2 MRI images of thighs during 1st episode of myalgia demonstrating signs of myositis in the left quadriceps muscle. (B) Coronal T2 MRI image of right foot during 2nd episode of myalgia demonstrating signs of myositis in the plantar region of the foot and concomitant plantar fasciitis. (C) Coronal T2 MRI image of the cervical region during 4th episode of myalgia confirming diffuse enlargement, hyperintensity, and enhancement of sternocleidomastoid muscles, compatible with bilateral myositis.

Treatment of FM is typically limited to anti-inflammatory drugs.<sup>1,2</sup> It is uncertain if steroid regimens should be considered in cases where inflammatory markers remain normal. Some immunosuppressors have been used in the literature, with no consistent outcome.<sup>1</sup> Long-term follow-up is suggested because of the potential, unconfirmed risk of PM/DM in these patients. Physicians should be careful when diagnosing FM so as not to misdiagnose the early showings of an underlying systemic disorder such as PM/DM or eosinophilic myositis. Migrating FM should be suspected in patients presenting with focal inflammation of one muscle group in the absence of systemic symptoms suggestive of alternative diagnoses.

To our knowledge, this is the first pediatric case of bilateral relapsing migrating FM. This is also the first case of sternocleidomastoid involvement in a child. Given the variety of clinical presentations and the association between FM and conditions such as IMIDs and neoplasia, we believe more research is required to elucidate the physiopathology of FM and its relation to other auto-immune and inflammatory disorders.

#### DISCLOSURES


The authors have no conflicts of interest to disclose.

#### STATEMENT OF AUTHORSHIP

LLH: role in data collection and manuscript redaction; JL: role in data collection and manuscript redaction; MPM: role in data collection and manuscript revision; JJB: role in data collection and manuscript revision; BT: role in data collection and manuscript revision; PM: role in data collection and manuscript revision, corresponding author.

*Lawrence Ledoux-Hutchinson*

*CHU Sainte-Justine Research Center, Montreal, QC, Canada*

*Jimmy Li* 

*CHU Sainte-Justine Research Center, Montreal, QC, Canada*

*Marie-Paule Morin*

*CHU Sainte-Justine Research Center, Montreal, QC, Canada*

*Immunology and Rheumatology Division, Department of Pediatrics, CHU Sainte-Justine, Montreal, QC, Canada*

*Jean Jacques De Bruycker*

*CHU Sainte-Justine Research Center, Montreal, QC, Canada*

*Immunology and Rheumatology Division, Department of Pediatrics, CHU Sainte-Justine, Montreal, QC, Canada*

*Bruce Tapiero*

*CHU Sainte-Justine Research Center, Montreal, QC, Canada*

*Division of Infectious Diseases, Department of Pediatrics, CHU Sainte-Justine, Montreal, QC, Canada*

*Philippe Major*

*CHU Sainte-Justine Research Center, Montreal, QC, Canada*

*Division of Neurology, Department of Neurosciences and Pediatrics, CHU Sainte-Justine, Montreal, QC, Canada*

*Correspondence to:* Philippe Major, MD, FRCPC, CHU Sainte-Justine Research Center, 3175 Chemin de la Côte-Sainte-Catherine, Montreal, QC, Canada, H3T 1C5. Email: [philippe.major@umontreal.ca](mailto:philippe.major@umontreal.ca)

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