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Atypical Presentation of Human Acute Muscular Sarcocystosis: *Sarcocystis Nesbitti* Confirmed on Molecular Testing

Authors' Contribution:
Study Design A
Data Collection B
Statistical Analysis C
Data Interpretation D
Manuscript Preparation E
Literature Search F
Funds Collection G

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Conflict of interest: None declared

Patient: Male, 51
Final Diagnosis: Acute muscular sarcocystosis
Symptoms: Fever • myalgia
Medication: —
Clinical Procedure: —
Specialty: Infcetious Diseases

Objective: Rare disease

Background: Acute muscular sarcocystosis (AMS) is one of a spectrum of diseases caused by the *Sarcocystis* parasite which infects humans in regions where it is endemic. Infections present with non-specific signs and symptoms and have been known to occur in clusters.

Case Report: A 51-year-old Vietnamese male presented to Tan Tock Seng Hospital, Singapore with 3 weeks of fever, urticarial rash, non-productive cough, and lower back pain. He had an extensive travel history prior to presentation. Magnetic resonance imaging (MRI) showed myositis involving the paravertebral and upper thigh muscles. The infection was confirmed on open muscle biopsy and *Sarcocystis nesbitti* was identified on molecular testing. The patient was treated with prednisone and methotrexate.

Conclusions: AMS must be considered in a patient with history of exposure to an endemic area. Diagnosis of the condition and identification of *S. nesbitti* as the causative organism will help to further study of this particular condition and guide treatment.

MeSH Keywords: Eosinophilia • Magnetic Resonance Imaging • Myositis • Sarcocystosis • Polymerase Chain Reaction

Full-text PDF: https://www.amjcaserep.com/abstract/index/idArt/913327

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Background

Sarcocystis is a protozoan genus of parasite that requires an intermediate and a definitive host to complete its life cycle. Humans are known to serve as the intermediate hosts for several strains [1]; the most common presentations are intestinal and muscular sarcocystosis.

Infections have been described in travelers to parts of Southeast Asia where the parasite is endemic [1]. Acute muscular sarcocystosis (AMS) occurs after sporocysts are ingested from contaminated water or food. Schizonts develop in the blood and then invade muscle, whereupon they form sarcocysts [1].

This infection has been described in several outbreaks in Malaysia [2], which is in close proximity to our medical center in Singapore. The prevalence of human muscular sarcocystosis has been quoted to be up to 21% from autopsy specimens in Malaysia [3]. This is in contrast to a prevalence of between 0% and 3.6% in Western countries [4]. There is no established epidemiological data on sarcocystosis in Singapore.

With this report, we described an index case of AMS in Singapore with an unusual distribution of myositis. It is uncommon for *Sarcocystis* infections to occur in isolation, with several previous outbreaks described in various locations in Malaysia [2,5–8].

Case Report

Our patient was a 51-year-old Vietnamese male who presented with a fever for 3 weeks, along with a non-productive cough. His travel history in the 6 months preceding his clinical symptoms was extensive, including Malaysia, Japan, France, Vietnam, and Thailand. There was no history of outdoor activity nor ingestion of raw or undercooked foods.

Physical examination revealed extensive urticaria, as well as tenderness of the lower back and calves.

Initial blood tests demonstrated peripheral eosinophilia and transaminitis. Serum creatinine kinase was markedly raised at over 1900 UI/L. These findings were compatible with eosinophilic myositis. Total white blood cell levels were not raised. However, erythrocyte sedimentation rate and C-reactive protein levels were mildly elevated at 71.6 mm/hr and 20 mg/L, respectively. Mild acute kidney injury was also present with a serum creatinine of 107 µmol/L. Serological tests for organisms, such as toxoplasma, strongyloides, legionella, and Leptospira, were negative.

Magnetic resonance imaging (MRI) of the lumbar spine and thighs (Figures 1A, 1B, 2A, 2B) was performed to evaluate the complaints of pain. A routine musculoskeletal protocol was implemented, using both T1 and T2 weighted sequences prior to and after contrast administration. Patchy edema with corresponding contrast enhancement was seen, in keeping with myositis of the paravertebral, thigh, and partially imaged calf muscles.

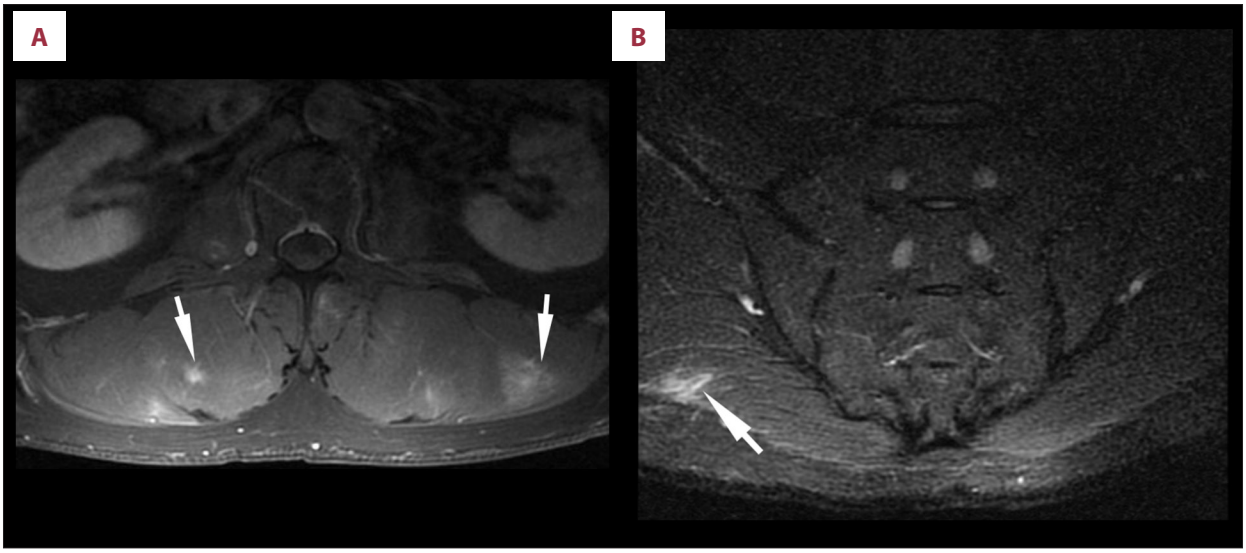

Figure 1. (**A**) T1-weighted fat-saturated post contrast sequence: axial image of the paraspinal muscles showing wispy contrast enhancement (white arrows) scattered throughout the muscles, in keeping with findings of myositis. (**B**) T1-weighted fat-saturated post contrast sequence: coronal image of the gluteus muscle showing a focus of enhancement (white arrow) in the right gluteus muscle.

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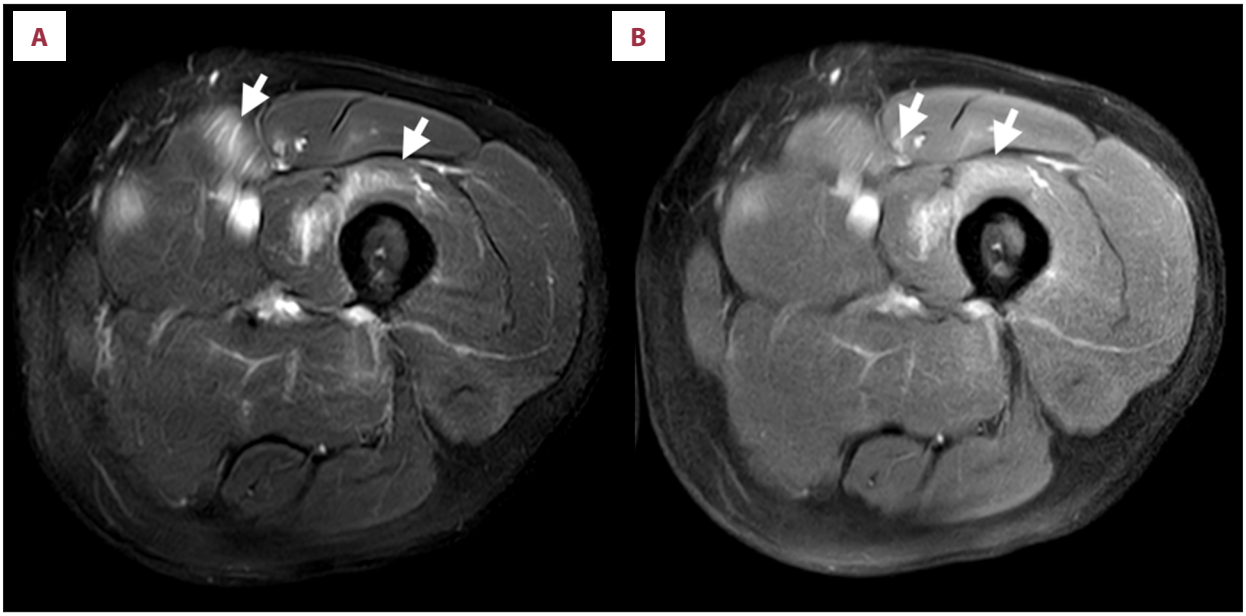

Figure 2. (**A**) T2-weighted fat-saturated sequence: axial image of the left thigh muscles showing areas of hyperintensity in the quadriceps and adductor muscles, in keeping with myositis. (**B**) T1-weighted fat-saturated post contrast sequence: coronal image of the left thigh muscles showing patchy enhancement corresponding with previously seen abnormal signal in the quadriceps and adductor muscles.

Open biopsy of the distal right vastus lateralis muscle showed inflammatory changes and the presence of sarcocysts. The patient was initially started on empirical treatment with albendazole and trimethoprim for parasitic infection, which was changed to prednisolone and co-trimoxazole upon receipt of the muscle biopsy histology. His myalgia improved only slightly on this regime and a polymerase chain reaction (PCR) of the biopsy sample was sent to the University of Malaya Department of Parasitology, which confirmed the presence of *Sarcocystis nesbitti*.

Methotrexate was then used in combination with prednisone. Serum creatinine kinase along with other abnormal serological markers continued to downtrend and normalize with treatment. His symptoms of calf and lower back pain frequently recurred, however, albeit less in intensity from his initial presentation, despite compliance to his treatment.

At present, our patient has returned to France and remains on methotrexate and prednisone. His slow recovery prompted his physicians there to repeat a muscle biopsy, which was performed 1 year after his initial presentation. Persistent myositis was demonstrated. No sarcocysts were seen in the sample.

Discussion

AMS is an eosinophilic myositis which has a wide spectrum of non-specific presenting symptoms such as fever, myalgia, muscle weakness, muscle swelling, headache, and cough [6], making early diagnosis and treatment potentially difficult [7]. Initial differential diagnoses were entertained in our patient case, such as vasculitis in view of the urticarial patches, as well as an underlying auto-immune disorder in view of fever and non-specific symptoms.

The disease in this case report followed a relatively well described initial course with symptoms that recurred despite treatment [8]. It has previously been observed that the duration of symptoms may last up to 24 months [1]. Persistent myositis at 1 year after commencement of treatment in our patient supported chronicity of inflammation, which was difficult to control even with steroids and immunosuppressants.

Involvement of the thigh muscle is atypical in AMS cases that have been described in the literature. Other documented sites of involvement have been the gastrocnemius, muscles of mastication, and superficial back muscles [2]. Abnormal signals in the thigh and calf musculature may be attributed to a wide variety of pathologies, including but not limited to auto-immune conditions, viral infections, and bacterial infections [9]. The utility of MRI in such conditions is to detect myositis, guiding muscle biopsies [2] to areas of active inflammation, which can appear as foci of contrast enhancement or hyperintensity on fluid weighted sequences.

Characteristic histologic visualization of sarcocysts within the muscle might be seen in as little as 40% of cases [7] and cannot

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identify the specific organism. Molecular testing with PCR to identify *S. nesbitti* as the culprit organism is becoming more important, as it has been detected in similar AMS cases in humans with greater frequency [7]. Knowledge of the behavior of *S. nesbitti* is key to prevention and treatment of this condition.

The definitive treatment of AMS is still under study [6]. The curative role of anti-parasitic agents, such as albendazole, requires additional study. Improvement in myalgia may be seen with steroids and immunosuppressants in some patients, such as in the case of our patient. Our patient remains on close outpatient follow-up in Singapore and France.

Conclusions

AMS is an important and under reported cause of myositis in patients with a history of exposure to an endemic area. Diagnosis of AMS and localization of *S. nesbitti* with imaging, histology, and molecular testing can help distinguish it from other potential causes of polymyositis and can help guide appropriate treatment and follow-up. Isolated cases such as this patient case contribute to the body of work on this subject, as we continue to study the potential sources of infection.

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