Non-tropical pyomiositis and severe subacute polyneuropathy

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Summary
Non-tropical pyomiositis is an uncommon disease, sometimes associated with other entities. Association with Guillain-Barré syndrome has been exceptionally reported. In this paper we present the case of a patient with non-tropical pyomiositis due to Escherichia coli who developed a subacute/acute demyelinating polyneuropathy. A 76-year-old man was admitted with muscle inflammation affecting the right shoulder, an abscess spontaneously draining in the right armpit, and a suppurative collection in the left forearm. After successful treatment he was discharged from the hospital, but 45 days later he developed subacute polyneuropathy. When admitted for this second time, a muscle abscess in the right forearm was observed and pus cultures were positive for Escherichia coli. With antibiotics according to antibiogram the patient improved, and after proper nutrition and intravenous immunoglobulins, he recovered slowly from polyneuropathy. This case shows the association of pyomyositis due to E. coli and acute demyelinating polyneuropathy. To our knowledge, only one similar case was reported; in that case the etiologic agent was Staphylococcus aureus. The association of the pyomiositis with the syndrome of Guillain-Barré is exceptional, and has been reported only in a patient affected by pyomiositis by Staphylococcus aureus. En el caso que aquí presentamos se cultivó una Escherichia coli en uno de los abscesos musculares que presentó el paciente, quien desarrolló una polineuropatía subaguda con disociación albúmino-citológica en el líquido cefalorraquídeo, compatible con síndrome de Guillain-Barré, por lo que constituye el primer caso de Guillain-Barré asociado a piomisitis por ese germe. Palabras clave: Piomiosis. Escherichia Coli. Guillain-Barré. Polineuropatía aguda. Absceso muscular

Key words: Pyomyositis. Escherichia Coli. Guillain-Barré. Acute polyneuropathy. Muscle abscess

Introduction
Pyomyositis is an uncommon condition, that mainly affects inhabitants from tropical areas [5], although in recent times an increasing number of patients from temperate zones has challenged such concept [3]. Classically, it is defined by the development of muscle abscesses/areas of pyomyositis, usually caused by Staphylococcus aureus, not related to endocarditis or any identifiable septic focus. A prompt diagnosis is important, since the disease may bear a fatal outcome [13]. Although it usually occurs as an isolated condition, sometimes it is associated with other entities, although the pathogenetic link remains obscure. One of these associations (very rarely reported) is Guillain-Barré syndrome [2]. The following case reports on a diabetic patient affected by pyomyositis who developed a subacute polyneuropathic syndrome. The germ isolated in this case was Escherichia coli. Infections due to Escherichia coli may be related to Guillain-Barré syndrome, but the association between Escherichia coli pyomyositis and Guillain-Barré syndrome has not been reported previously.
Presentation of case
A 76 year-old male patient was admitted to our unit after having been found by a family member after having fallen at home, being unable to stand up without aid. The patient had been witnessed by another family member 4 hours before, without any apparent problem. Previous diagnoses included diabetes mellitus and arterial hypertension. The patient also received acetyl-salicylic acid 100 mg per day, betahistine, insulin, lercanidipine, losartan, metformine and simvastatine.

At admission the patient presented multiple chest, abdomen and knees contusions; a draining abscess located in the right armpit was also identified, as well as a localised swelling at the left forearm, that spontaneously drained, a couple of days later, purulent material and resolved. He complained of pain affecting the right shoulder and right arm. The patient did not refer aggression/violence, only that he fell down and suffered a nearly loss of consciousness. The remaining physical examination was irrelevant including the absence of cardiac murmurs.

Figure 1. Coronal STIR, showing intense signal affecting the right supraspinatus and infraspinatus muscles (arrows). This lesion is highly suggestive of oedema/inflammation of the affected muscles.
Laboratory evaluation at admission was consistent with rhabdomyolysis, with raised serum creatinphosphokinase (CPK) 13 759 U/L, serum creatinine 2.7 mg/dL, aspartate aminotransferase (ASAT) 474 U/L, alanine aminotransferase (ALAT) 186 U/L, lactate dehydrogenase (LDH) 2499 U/L. Hematuria ++++. An echocardiography was negative for endocarditis, and blood cultures were negative; also, brain computed tomography scan (CT) and electroencephalogram (EEG) were both normal.

The patient received intravenous fluids and antibiotics, and the acute rhabdomyolysis subsided. However, pain and impaired mobility of the right arm persisted. For this reason, he underwent magnetic resonance imaging (MRI) of the cervical area and brachial plexus, that was consistent with muscle oedema/inflammation, especially affecting supraspinatus and infraspinatus muscles (Fig.1).

After a few days of treatment with levofloxacin and ceftriaxone, the general status of the patient improved and he regained the ability to walk. The patient was remitted to a medical facility, where he remained for two months.

After this period, the patient was admitted again to this hospital, complaining of severe weakness affecting both legs and both arms, that had begun 15-20 days before and progressively increased, together with pain and localised right forearm swelling. Neurophysiologic exploration revealed severe, acute, sensitive-motor axonal polyneuropathy. The patient denied any contact with heavy metals or other toxic compounds. Neoplasia was excluded after performing a whole-body CT scan, as well as autoimmune disease (negative ANA and ANCA determinations). Antiganglioside antibodies were also negative. No skin or liver abnormalities were detected. Lumbar puncture allowed extraction of transparent fluid (CSF) with 20 leucocytes and >300 mg/dl proteins. Although the patient reported that weakness had begun at least 2-3 weeks before, treatment with immunoglobulins was administered, with slightly clinical significant improvement. However, two lumbar punctures performed during the following weeks showed a progressive decrease in both cells (2 leucocytes) and proteins (126 mg/dl). Due to severe malnutrition and reduced ability to eat, percutaneous endoscopic gastrostomy was performed. During the hospital stay, a progressive increase of the right forearm swelling was observed, accompanied by raised acute phase reactants and fever. A MRI showed an image compatible with abscess (Fig.2) that was drained obtaining a purulent fluid in which *Escherichia coli* was isolated. The patient was treated with meropenem and amikacin according to the results of antibiogram. Echocardiography was negative. Local symptoms disappeared and the general status improved slowly. No other episode of muscle inflammation has been identified since then.

**Discussion**

There are several striking clinical features that concur in this patient. At first, the presence of several purulent collections, two of them spontaneously draining (right armpit and left forearm), and another one filled with purulent fluid that grew *E. coli*. The second striking feature is the development of a severe subacute/acute polyneuropathy accompanied by increased proteins

Figure 2. Sagital T1 with contrast showing a ring-like enhancement of volar muscles of the proximal third of the right forearm, in relation to an abscess (arrowhead).
Pyomyositis is a frequently overlooked disease. The most commonly isolated microorganism in cases of tropical pyomyositis is *Staphylococcus aureus* (75-90% of cases), but other germs have been involved [4,7]. *Escherichia coli* pyomyositis has also been described [6,14], as in the present case. Muscle infarcts and myonecrosis have also been reported in diabetes [12], but MRI features do not support this diagnosis, but that of pyomyositis.

On the other hand, the patient developed a severe subacute polyneuropathy, that caused tetraparesia and marked muscle atrophy. The etiology of this neural affection is obscure. Paraneoplastic, toxic, infectious and autoimmune causes were reasonably discarded, and the patient had no features or history of porphyria. The Guillain-Barre syndrome is a possibility, although the evolution of the symptoms –although well described in patients with Guillain-Barré- is unusual for this entity. The patient was diabetic, but the evolution of diabetes-associated polyneuropathy is not as acute as in this case. Another important entity in the differential diagnosis would be critical illness associated polyneuropathy, but the patient was doing well after the first admission and the two suppurative processes he presented cured with antibiotics and spontaneous drainage. So, of the mentioned etiologic possibilities, the most probable one is the Guillain-Barré syndrome; the increased CSF protein, the paucity of cells, as well as the improvement with immunoglobulins support this diagnosis. Interestingly, Bhargava et al.[2] reported the case of a patient affected by tropical pyomyositis who also presented Guillain-Barré syndrome. Whether or not this represents a casual association or really constitutes a complication of pyomyositis is unknown. In the reported case by Bhargava et al., the causative agent of pyomyositis was *Staphylococcus aureus* and not *E. coli* as in this case. Bhargava et al. support the possible relationship between Guillain-Barré syndrome and staphylococcal pyomyositis because of the existence of already reported cases of endocarditis due to *Staphylococcus aureus* and Guillain-Barré syndrome[1], raising the possibility of molecular mimicry and autoimmune reaction. However, *Escherichia coli* has been also involved in the development of a Guillain-Barré syndrome [8-10].

**Conclusion**

In conclusion we presented a case of a patient with multiple muscle abscesses that appeared successively within five months. In one of them *E.coli* was isolated that was successfully treated with meropenem and amikacine. No endocarditis was detected and the abscesses affected distant muscles, a clinical picture fully compatible with pyomyositis. The development of the subacute polyradiculoneuritis appeared 2-3 months after the first abscesses, raising the possibility that both entities are related to each other, in accordance with some other exceptional cases reported in the medical literature [8].

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**Competing interest**

Authors declare that there are no conflicts of interest regarding this case.

**Bibliography**