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Case report

Escherichia coli pyomyositis in a patient with Down syndrome: A case report

Keisuke Fujioka[a,*], Shunsuke Takeuchi[a], Takahiro Tayama[a], Mikiko Takei[a], Akemi Ono[a], Miki Shono[a], Koichi Shichijo[a], Tsutomu Narita[b], Shuji Kondo[a]

[a] *Department of Pediatrics, Tokushima Prefectural Central Hospital, Kuramoto-cho 1-10-3, Tokushima, Tokushima 770-8539, Japan*
[b] *Narita Child Clinic, Tenno-cho 32-9, Takatsuki, Osaka 569-0088, Japan*

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ABSTRACT

Pyomyositis is an infection of the skeletal muscle that involves intramuscular abscess formation. It is typically caused by gram-positive bacteria, especially *Staphylococcus aureus*. Few cases of *Escherichia coli* pyomyositis have been reported in immunocompromised adult patients, while none have been reported in children.

We present a case of a 4-year-old boy with Down syndrome who developed *Escherichia coli* pyomyositis. The patient presented to our hospital with a fever and right forearm swelling. The magnetic resonance imaging findings suggested pyomyositis of the right forearm muscle and osteomyelitis of the distal radius. Both the blood and puncture fluid cultures were negative. Cefazolin and vancomycin were administered, and his blood examination results and right forearm swelling improved; however, a slight fever persisted. The multiplex polymerase chain reaction isolated the *chuA* gene but not the *YjaA* gene; thus the patient was diagnosed with pyomyositis and osteomyelitis caused by *Escherichia coli* group D. The cefazolin was substituted with meropenem, and the vancomycin was discontinued. Thereafter, his fever promptly improved, which indicated that the cause of persistent fever was vancomycin drug fever. The patient was discharged after receiving 3 weeks of intravenous antimicrobial therapy, and recovered fully with no long-term sequelae.

To the best of our knowledge, this is the first reported case of *Escherichia coli* pyomyositis in a child. The findings in this case suggest that *Escherichia coli* should be considered when choosing initial empiric therapy for pyomyositis, especially in children with underlying conditions.

Introduction

Pyomyositis is defined as a primary pyogenic infection of the striated skeletal muscle. *Staphylococcus aureus* is the most common causative agent; it is responsible for up to 50 %_95 % of pyomyositis cases in all age groups [1]. It is uncommon for gram-negative organisms to cause pyomyositis. [2]. There are only a few reports of pyomyositis caused by *Escherichia coli*. These cases mostly occurred in immunosuppressed or chronically ill adult patients, but not in children. Herein, we report on a case of *Escherichia coli* pyomyositis in a patient with Down syndrome.

Case report

A 4-year-old boy with Down syndrome presented to our hospital with a fever and right forearm swelling. The patient had no relevant medical history such as severe infection. His mother did not recall the child experiencing any physical trauma. Blood examination found an elevated white blood cell (WBC) count and C-reactive protein (CRP); 13,600/μl and 9.6 mg/dl, respectively. Ultrasonography showed swollen subcutaneous tissue and a hypoechoic lesion between the antebrachial muscles. The hypoechoic lesion seemed to represent abscess formation, and cefazolin (50 mg/kg, q8h) and vancomycin (15 mg/kg, q6h) were administered while awaiting further examination through magnetic resonance imaging (MRI). Three days after being admitted, the T2-weighted short-tau inversion recovery and T1-weighted MRIs revealed high-intensity lesion around the antebrachial muscles and a low-intensity area at the distal radius, respectively (Fig.1). Part of the abnormal signal intensity around the antebrachial

* Corresponding author.
E-mail address: sarcae315@gmail.com (K. Fujioka).

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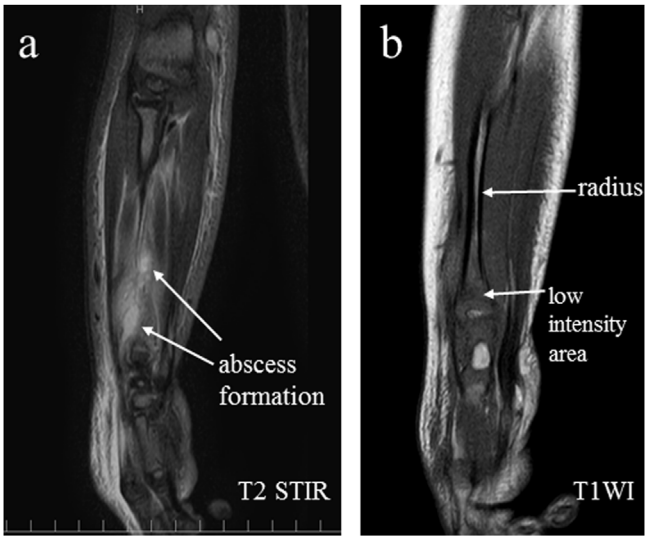

Fig. 1. MRI findings. **a** High intensity lesion around antebrachial muscles on T2 STIR implied abscess formation. **b** Low intensity lesion at distal radius on T1WI indicated osteomyelitis.

muscles suggested abscess formation. The patient was diagnosed with pyomyositis and osteomyelitis, and aspiration and drainage were performed on the same day. The forearm swelling gradually decreased, and the blood examination results also improved; however, a slight fever persisted. Both the blood and puncture fluid cultures were negative, and a polymerase chain reaction (PCR) test was performed to further investigate the etiology. Multiplex PCR isolated the *chuA* gene without the *YjaA* gene, which implied pyomyositis and osteomyelitis caused by *Escherichia coli* group D. The vancomycin was discontinued, and cefazolin was substituted with meropenem (40 mg/kg, q8h) considering extended-spectrum β-lactamase producing *Escherichia coli* (Fig.2). After withdrawing the vancomycin, his fever resolved immediately; thus, it was concluded that his persistent fever was due to vancomycin drug fever. On 3 weeks of admission, MRI showed disappearance of the high intensity lesion in the forearm, and the serum CRP level decreased to within the normal range. The patient was discharged on a 3-week course of oral trimethoprim-sulfamethoxazole (8 mg/kg of trimethoprim plus 40 mg/kg of sulfamethoxazole). He recovered fully, with no long-term sequelae.

Discussion

Pyomyositis is an infection of the skeletal muscle that involves the formation of intramuscular abscesses. It often occurs after an acute upper respiratory infection [3]. It is hypothesized that trauma to the muscle is required for deep tissue infection to occur. However, a history of trauma has only been elicited in 27 % of cases [1]. There was neither an antecedent infection nor a history of trauma in this case; therefore, the route of infection was unknown; however, we hypothesized that the skin infection was secondary to a bloodstream infection. Pyomyositis due to *Escherichia coli* is quite rare, and it seems unnatural that enteric gram-negative rods could occur in a cutaneous infection without an episode of trauma episode.

Staphylococcus aureus is the most common causative agent for pyomyositis; it is responsible for up to 50 %_95 % of cases in all age groups [1]. It is uncommon for pyomyositis to be caused by a gram-negative organism [2]. There are only a few reports of pyomyositis caused by *Escherichia coli*. These mostly occur in immunosuppressed or chronically ill adult patients, such as those with hematologic malignancies [4]. Vigil et al. reported 6 cases of *Escherichia coli* pyomyositis, all of which were receiving chemotherapy [4]. Another previous report showed that pyomyositis due to *Escherichia coli* occurred in HIV infection and diabetes [5]. To the best of our knowledge, this is the first report on a case of *Escherichia coli* pyomyositis in a child. It was estimated that mortality rate of *Escherichia coli* pyomyositis was higher than that of *Escherichia coli* blood stream infection [4]. Pyomyositis is difficult to diagnose at an early stage because of the subtle clinical presentation, which may lead to the poor prognosis.

It has been reported that almost all cases of *Escherichia coli* pyomyositis were caused by group B2, which was the main group found in a prospective study aimed at characterizing the risk factors for *Escherichia coli* bacteremia [6]. In this case, the *Escherichia coli* that was isolated belonged to group D. To our

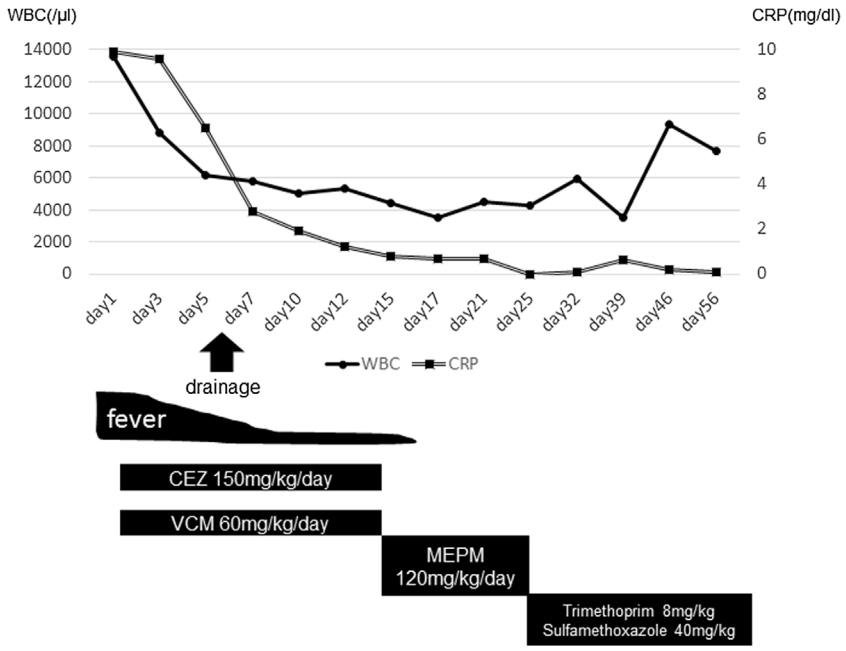

Fig. 2. Clinical course.

knowledge, only one case of pyomyositis caused by *Escherichia coli* group D has been reported on [3]. Studies have found that group B2 usually carries more virulence-associated genes than other phylogenetic groups [6]. Pyomyositis that was caused by group D, and not group B2 may have led to the good prognosis in this case.

Down syndrome is the most common chromosomal condition. It is associated with intellectual disability and is characterized by a variety of additional clinical findings. A third copy of chromosome 21, trisomy 21, has long been recognized as the cause of Down syndrome. It is known that increased susceptibility to infections is a feature of Down syndrome. Specifically, pneumonia is a major cause of morbidity and mortality in this patient population [7,8]. Regarding the immune system abnormalities, studies in the 1970s reported that Down syndrome is associated with neutrophil chemotaxis [9]. Moreover, the serum immunoglobulin (Ig) G1 and IgG3 concentrations are significantly increased in children with Down syndrome; however, the IgG2 and IgG4 concentrations are reduced [8]. This patient had no history of severe infection; however, the patient may have been susceptible to infection. After discharge, his immunocompetence was investigated, and his IgG subclasses and CD 4/8 ratio were found to be within the normal ranges.

In summary, here, we report on a case of *Escherichia coli* pyomyositis in a patient with Down syndrome. To the best of our knowledge, this case is the first to report on *Escherichia coli* pyomyositis in a child. Pyomyositis due to *Escherichia coli* is very rare and it mostly occurs in immunocompromised patients. Although studies have shown that the clinical course of *Escherichia coli* myositis can be severe, this patient recovered fully with no long-term sequelae. This case suggests that *Escherichia coli* should be considered when making decisions on the initial empiric therapy for pyomyositis, especially in immunocompromised patients.

Conflicts of interest

The authors declare no conflicts of interest associated with this manuscript

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Consent

Written informed consent was obtained from the parents of patient for publication of this case report and any accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Ethical approval

This case report has been granted an exemption from requiring ethics approval with the hospital-based comprehensive informed consent according to the Ethics Board of Tokushima Prefectural Central Hospital Japan

Author contribution

KF is the main author and wrote the manuscript. ST, TT, MT, AO, MS and KS were responsible for the diagnosis and treatment of the patient. TN gave technical support. SK critically reviewed the manuscript and supervised the study process. All authors read and approved the final manuscript

CRediT authorship contribution statement

Keisuke Fujioka: Conceptualization, Methodology, Writing - original draft. **Shunsuke Takeuchi:** Software, Investigation. **Takahiro Tayama:** Validation. **Mikiko Takei:** Investigation. **Akemi Ono:** Investigation, Data curation. **Miki Shono:** Data curation. **Koichi Shichijo:** Software, Validation. **Tsutomu Narita:** Supervision, Data curation. **Shuji Kondo:** Writing - review & editing.

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