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Case Report

The First Report of Septic Arthritis of the Pubic Symphysis Associated With Fournier's Gangrene

Osamu Okamoto[1*], Seiichi Sato[2], Kaho Matsuda[2], Noriko Gamachi[3], Ken-ichi Hirai[4], and Hiroyuki Hashimoto[2]

[1]*Dermatology, Oita City Medical Association's Almeida Memorial Hospital*

[2]*Plastic surgery, Oita City Medical Association's Almeida Memorial Hospital*

[3]*Clinical pathology, Oita City Medical Association's Almeida Memorial Hospital*

[4]*Urology, Oita City Medical Association's Almeida Memorial Hospital*

**Corresponding author: Dr. Osamu Okamoto MD, PhD, Oita City Medical Association's Almeida Memorial Hospital, 1509-2, Miyazaki, Oita, 870-1195, Tel.:+81 97 569 3121; Fax: +81 97 569 9746, Email: ookamoto@ oita-u.ac.jp*

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Abstract

A 66-year-old male was diagnosed as Fournier's gangrene with 6% skin involvement. The patient underwent debridement followed by the placement of a skin graft. After the completion of the treatment, a small granulomatous ulcer with an abscess remained in the pubic area. A wide excision of the abscess was performed, revealing the presence of necrotic tissue in the pubic symphysis; however, no evidence of osteomyeliitis was observed. The excised tissue revealed necrotic cartilage, and enterococcus was detected. A diagnosis of septic arthritis of the pubic symphysis was made. CT studies were useful for detecting changes in the pubic symphysis. This is the first report of septic arthritis of the pubic symphysis as a complication of Fournier's gangrene. Septic arthritis of the pubic symphysis is a rare disease in the orthopedic and gynecological field; therefore the present case supplies useful information for dermatologists for the examination of non-healing ulcers in the pubic area.

Keywords: Fournier's Gangrene; Septic Arthritis; Pubic Symphysis; Non-Healing Ulcer; Cartilage Necrosis

Introduction

The infection of the pubic symphysis is called septic arthritis of the pubic symphysis (SAPS) [1] which is a rare disease in the orthopedic and gynecological field. The major cause of SAPS, which most commonly occurs in athletes, is repetitive traction stimuli of the pubic symphysis because several adductor muscles are attached to the pubic symphysis [1,2]. SAPS are also known to occur in women during the postpartum period and women who undergo surgery to treat urinary incontinence [3]. Apart from these major causes, there are occasional reports of other causes, including pelvic malignancies and drug abuse [1,4]. Causative bacteria are various; among them, *Staphylococcus aureus* predominates, followed by *Pseudomonas aeruginosa* [1]. It is believed that such infections occur either during surgery or as a result of pelvic malignancy. On the other hand, for athletes, it is estimated that a microtrauma forms in the pubic symphysis due to repetitive traction stimuli, then, when occasional bacteremia occurs, the bacteria colonize in the trauma [5].

Fournier's gangrene is a form of necrotic fasciitis that occurs due to a bacterial infection in the genital area [6]. The infection extends widely under the skin and the clinical symptoms tend to become severe [7]. Although the lesion involves the pubic area, there are no reports of

SAPS occurring in association with Fournier's gangrene.

We experienced a case of Fournier's gangrene which presented with a widespread lesion [8]. Most of the lesions were covered by grafted skin however; a small ulcer with edematous granulation tissue remained and persisted for several months. After several additional operations, the patient was diagnosed as SAPS. We herein report an unusual case of SAPS as a complication of Fournier's gangrene. Because dermatologists are not familiar with the condition, this report is helpful for them for examining non-healing ulcers in the pubic area.

Case

The episode of Fournier's gangrene of the present case has previously been reported [8]. Briefly, a 66-year old male admitted to our hospital in March due to a painful swelling of the left genital area. He was diagnosed as Fournier's gangrene, with lesions that extended from the lower abdomen to the left thigh. Methicillin-resistant *Staphylococcus aureus* (MRSA), *Peptostreptococcus* species, and *Enterococcus faecalis* were successively detected in the pus from the pubic area during April and May.

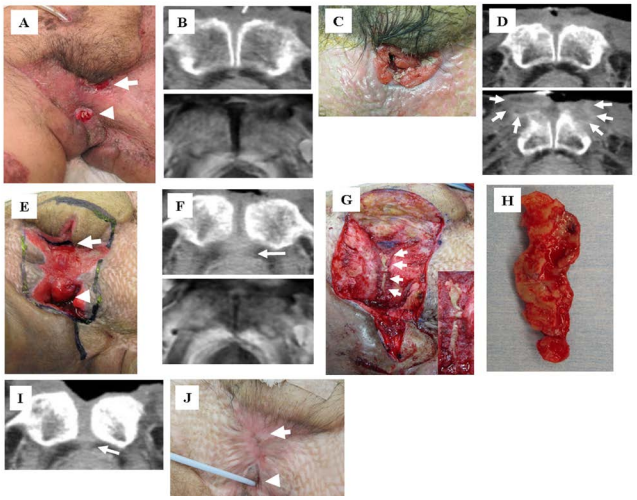

Figure 1. Summary of the clinical course. A: The initial appearance of the ulcers in July. Arrow: a non-healing ulcer proximal to the pubic bone. Arrowhead: an ulcer corresponding to the edge of the amputated penis. B: A CT image (upper panel) and an MRI image (lower panel) taken at a similar time, respectively. C: A close-up view of the ulcer proximal to the pubic bone in September. D: Upper panel: A CT image showing a low-density area of the pubic symphysis. Lower panel: A CT image of the abscess (arrows). E: The design of the excision. F: A CT image (upper panel). The connection between the pubic bone and the cavernous body is indicated by an arrow. Lower panel: An MRI image taken at a similar time. G: The direct observation of the pubic symphysis. White necrotic tissue is indicated by arrows. Inset: A close-up view of the necrotic tissue. H: The excised necrotic tissue. I: A CT image one month after the excision. The connection between the pubic bone and the cavernous body is indicated by an arrow. J: Appearance of the pubic area six months after removal of the necrotic cartilage. An urethral fistula is catheterized at this time. In E and J, the arrows and arrowheads correspond to those in panel A.

Episodes of sepsis (a fever and highly elevated CRP and procalcitonin levels) were observed in the same period. The patient underwent debridement, penoscrotectomy, and the construction of a colorectal stoma in early May. The lesions were then covered with a skin graft in late May and the patient was discharged in June.

After the above treatments, a small ulcer just above the pubic area from which the discharge of ill-odored pus, was observed. Another ulcer, which corresponded to an edge of the penile amputation also remained (Figure1A). In September, the formation of edematous granulation tissue was observed to have increased in the former ulcer (Figure1C) and a CT examination revealed an abscess just above the pubic symphysis (Figure1D, bottom panel). Repeated debridement did not improve the symptom and finally, two fistulae developed in the proximal and distal sides of the pubic bone (Figure. 1E). Vancomycin was administered after MRSA was detected from the pus.

In CT examinations, a low-density area appeared in the pubic symphysis (compare Figure1B, and D, F). The pubic symphysis became widened (Figure1F), and a soft tissue shadow connected the pubic bone and the remaining cavernous body of the penis (Figure1F). However, repeated MRI examinations did not indicate the presence of osteomyelitis (Figure1B, F). Because we suspected an infection in the pubic symphysis, a wide local excision of the ulcer was performed in December (Figure1E).

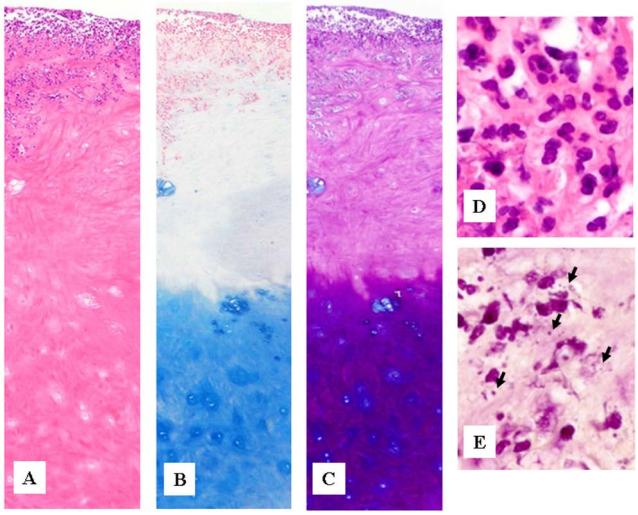

Figure 2. Histological findings of the excised necrotic tissue. A: H-E staining. B: Alucian blue staining. Viable cartilage is stained blue. C: Double staining of alucian blue and PAS. PAS stains necrotic cartilage in pink. A violet color indicates the merging of pink due to PAS and blue due to alucian blue. A-C: Original magnification X40. D: A higher magnification of infiltrating cells (H-E staining). E: A higher magnification of the cocci by Gram staining (arrows). D and E: Original magnification X400.

After the removal of the granulation tissue on the pubic bone, a white, linear necrotic tissue was found in the pubic symphysis (Figure1G). The necrotic tissue occupied half of the volume of the pubic symphysis (Figure1H). The partial removal of the cortex of the pubic bone revealed good

bleeding; osteomyelitis was not observed. *Enterococcus* spp. was cultured from the excised tissue. According to these findings, a diagnosis of SAPS was made. One month after the operation, in a CT examination, the symphysis became wider, but the low-density area disappeared and the connection between the pubic bone and the cavernous body of the penis decreased in size, (Figure1I). The ulcer proximal to the pubic bone healed several months later and no recurrence of the fistula was observed (Figure1J). Another ulcer, which was later revealed to be a urethral fistula, was managed by catheterization and it finally closed.

A histological analysis revealed that the excised necrotic tissue was cartilage with inflammatory cell infiltration (Figure 2). On the tissue surface, the chondrocytes disappeared and the area became alucian blue-negative and PAS-positive, indicating necrosis (Figure 2B, C). Many neutrophils and Gram-positive cocci were observed in the surface (Figure 2D, E).

Discussion

SAPS is a rare infectious disease of the pubic joint [1,9]. Pubic pain, waddling gait, and pain during hip movement are common symptoms, and general fever, bacteremia, leukocytosis, osteomyelitis are common findings. The patient in the present case had been bedridden for an extended period of time and did not complain of pain during movement. Instead the patient presented with non-healing ulcers that showed the sustained discharge of pus. SAPS was eventually diagnosed after we denuded the granulation tissue on the pubic bone and found a necrotic cartilage. The present case suggests the benefit of performing a direct examination of the pubic symphysis when a non-healing ulcer is encountered in the pubic region.

The pubic region is usually involved in Fournier's gangrene; however, there have been no previous reports of SAPS in association with Fournier's gangrene. Furthermore, there are few reports which describe the histology of the affected cartilage in cases of SAPS; thus, the microscopic findings of the present case may be helpful for broadening the understanding of the pathology of this condition. The necrosis of the cartilage is thought to have been induced by bacterial infection. Accordingly, necrosis on the surface of the cartilage and bacterial colonization were both proven, and neutrophilic infiltration was observed. Nevertheless a tissue culture revealed only a small amount of enterococcus – this was likely the result of the administration of vancomycin before the tissue was cultured. Regarding origins of the bacteria, both direct infection from an abscess due to Fournier's gangrene and indirect infection during septic episodes are possible. However, due to surface infection of the cartilage, the former mechanism would be more feasible.

MRI is known to be more useful than CT for the detection of SAPS [10]. Recent publications have demonstrated SAPS lesions as clear areas of high-intensity [4,9]. In the present case, we did not observe an abnormality of intensity; it should therefore be noted that SAPS lesions cannot always be detected by MRI. On the other hand, low-density areas were detected within the pubic symphysis by CT imaging. In addition to the density change, the widening of the pubic symphysis was a characteristic finding. This finding is reportedly seen in about 30% of the SAPS cases [1], and proceeded according to the clinical course.

We reported the first case of SAPS associated with Fournier's gangrene. SAPS was identified based on the presence of non-healing ulcers after surgery, and necrosis of pubic symphyseal cartilage was identified as a cause of the non-preferred clinical course. SAPS is mainly managed by gynecologists and orthopedic surgeons, and this disease is not familiar to dermatologists. Therefore similar SAPS cases might go unnoticed in the dermatological field. The present case would help dermatologists to imagine the presence of SAPS when they encounter a non-healing ulcer in the pubic area.

Conflict of interest

The authors have no conflict of interest to declare.

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