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Endovascular repair of a Clostridium perfringens infected pseudoaneurysm presenting as an intramural air pocket

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ABSTRACT

An infected aneurysm (IA) is a relatively rare but complex and life-threatening disease. We report a 78year-old man with an IA in the common iliac artery (CIA) due to *Clostridium perfringens*. An initial computed tomography (CT) revealed an air pocket in the left CIA, and a pseudoaneurysm was seen on the CT taken the next day, in the area where the air pocket was initially observed. Due to the patient's high surgical risk, emergent endovascular aneurysm repair (EVAR) was performed. No indolent infection was found 1.5 years after the surgery. Because of its high risk of expansion and rupture, accurate diagnosis and immediate treatment is required for managing IAs. The case emphasizes that air density in an arterial wall could be an early radiologic feature of an IA, and EVAR could be a treatment option for IA. © 2020 The Author(s). Published by Elsevier Ltd. This is an open access article under the CC BY-NC-ND license (http://creativecommons.org/licenses/by-nc-nd/4.0/).

An infected aneurysm (IA) is a relatively rare but lifethreatening condition. Because of its rapid progression and high mortality rates, accurate diagnosis and immediate treatment is essential. Open surgical repair with resection of the infected tissue, aggressive debridement, and in situ graft replacement or extraanatomical bypass is considered to be the standard treatment. However, operative intervention is associated with high morbidity and mortality rates. Recently, emergent endovascular aneurysm repair (EVAR) has become one of the treatment approaches for IAs. We observed the process of IA formation by consecutive computed tomography (CT) scanning which could aid in early diagnosis.

CASE REPORT

A 78-year-old man was admitted to our hospital due to fever and quadriplegia. He had a history of chronic heart failure with low ejection fraction of about 20%, coronary artery bypass grafting, percutaneous coronary intervention, hyperlipidemia, and chronic kidney disease. On the day of admission, his temperature was 38.3°C. The laboratory findings included WBC 18,800/μL, creatinine 1.99 mg/dL and CRP 30.36 mg/dL. Plain CT revealed an air pocket in the left common iliac artery (CIA) (Fig. 1). A dynamic CT taken the next day showed pseudoaneurysm of the left CIA with panniculitis (Fig. 2). As he was a high-risk surgical patient,

* Corresponding author at: Department of Surgery, Tokyo Dental College Ichikawa General Hospital, 5-11-13 Sugano, Ichikawa, Chiba, Japan. *E-mail address:* sono@tdc.ac.jp (S. Ono). conservative antibacterial therapy with tazobactam/piperacillin (TAZ/PIPC) was sustained. On day 7 of hospitalization, *Clostridium perfringens (C. perfringens)*, which was sensitive to TAZ/PIPC, was reported in the blood culture taken on the day of admission. Because of increasing lumbar pain, a CT scan was taken on day 8 of hospitalization which revealed rapid growth of the pseudoaneurysm (Fig. 3). Due to the patient's high surgical risk, emergent EVAR and left internal iliac artery embolism were performed under spinal anesthesia.

His postoperative course was uneventful. A CT taken 7 days after the operation revealed exclusion of the pseudoaneurysm (Fig. 4). Intravenous antibacterial treatment was administered for 14 days, and he was discharged on postoperative day 16 with oral amoxicillin/clavulanic acid for 6 months. However, the patient defaulted his appointment and antibacterial treatment. In fact, it turned out that he took oral antibacterials for only about 14 days.

One and half years after the surgery, he happened to visit our hospital with other symptoms, and CT revealed no evidence of indolent infection (Fig. 5).

DISCUSSION

IAs are estimated to account for 1-2% of all aortic aneurysms and are well-known as complex and life-threatening diseases because of their high risk of expansion and rupture [1].

Clinical manifestations include fever and pain associated with leukocytosis and increased C-reactive protein levels. In our case, non-contrast CT identified an air pocket in the wall of the left CIA,

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Fig. 1. The arrow indicates an air pocket in the left common iliac artery.



Fig. 2. The arrow indicates a pseudoaneurysm in the left common iliac artery with panniculitis.

and follow-up enhanced CT revealed a pseudoaneurysm where the air pocket had been recognized. The diagnosis of an IA due to the presence of an intra-wall air pocket before the formation of an aneurysm is extremely rare. Reviewing the course, we could diagnose even before the formation of the aneurysm by an air pocket detected by CT. Further cases should be observed to understand the progression of IAs.

Infected abdominal aortic aneurysm (IAAA) is mainly caused by *Salmonella* or *Staphylococcus*, but other pathogens have also been identified [2]. Blood cultures are negative in 25-50% of cases, and the primary cause of sepsis is not identified in approximately one third of the patients [1]. In many cases, the infectious etiology can be isolated only after removal and culture of the involved tissue.

C. perfringens is a Gram-positive spore-forming anaerobic bacillus. It produces an alpha-toxin that acts as a phospholipase, degrading tissue and cell membranes. The organism can be associated with hemolysis and myonecrosis, commonly known as



Fig. 3. The arrow indicates the rapidly growing pseudoaneurysm.



Fig. 4. The arrow indicates the exclusion of the pseudoaneurysm.

gas gangrene. *C. perfringens* is ever-present in nature and can be found as a normal component of decaying vegetation and marine sediment, in the intestinal tract of humans and other vertebrates and insects, and in the soil. It typically clinically manifests in deep penetrating traumatic wounds with vascular compromise. This report describes a common iliac artery IA whose cultures were positive for *C. perfringens* alone [3], an unusual isolate in this type of infection. Sherwood et al. reported a review of the literature that showed only two similar previous cases from cultures of intraluminal thrombus [3,4]. In our case, the patient had no traumatic events or digestive symptoms. It still remains unclear how he was infected with the bacterium.

The conventional treatment for IAAA is open surgical repair (OSR) combined with antibacterial therapy. Examples of OSR procedures include surgical resection, debridement of infected periaortic tissues, and reconstruction of the aortic flow. However, despite advances in perioperative management, OSR remains



Fig. 5. The computed tomography scan 1.5 years after the surgery shows no evidence of indolent infection.

associated with high morbidity and mortality rates [2]. The short-term mortality rate after OSR is as high as 31-44% [5–8].

In our case, we diagnosed IA from the laboratory data and the CT findings which revealed an air pocket in the left CIA. Generally, OSR is considered to be a standard strategy. However, due to the high surgical risk of the patient, emergent EVAR under spinal anesthesia was performed. EVAR was first applied to IAs in the 1990s, when it had only recently emerged [9]. Since then, it has been described as an alternative to OSR [10–13]. Kritpracha et al. used EVAR to treat 16 patients with IAAA who were without aortoenteric fistulas (AEF) and reported an in-hospital mortality rate of 6% [14]. Xiao et al. used EVAR to treat 19 patients with IAAA. They reported a one-year survival rate of 73.7%, and the 2-year and 5-year survival rates were both 64.9% [2]. Thus, it appears that EVAR combined with sufficiently prolonged antibacterial treatment is a feasible alternative to OSR for IAAA. However, there are detriments to this strategy. A major disadvantage of EVAR is the lack of resection of the infected tissue, which is the essential part of OSR. The remaining infected tissue and fluid can facilitate recurrent infection and sepsis, leading to catastrophic events. For those reasons, OSR is the first treatment option for IAAA, and EVAR is commonly the only therapeutic option for patients with high surgical risk.

Duration of the postoperative antibacterial therapy is crucial [13,15,16]. Although administration for a minimum of 1–2 months is recommended by some authors, administration for 6 months or for the rest of the patient's life is claimed to be required in more recent reports [11,13,16]. In our case, antibacterial treatment was continued for only 14 days after administration. It would be a high risk for indolent infections; however, no such event has occurred one and half years after the surgery.

CONCLUSION

We observed a rare case which suggests that the course of aneurysm formation can be followed by consecutive CT. An air pocket in an arterial wall could be an early radiologic feature of an IA, and EVAR could be one of the feasible alternatives for managing IAs.

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Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images.

Declaration of Competing Interest

The authors have no conflicts of interest to declare.

CRediT authorship contribution statement

Takayuki Tsuji: Writing - original draft. Shigeshi Ono: Writing - review & editing, Supervision. Keisuke Eguchi: Writing - review & editing. Noriaki Wada: Writing - review & editing. Hirotoshi Hasegawa: Writing - review & editing. Junichi Matsui: Writing review & editing.

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