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Case report

Osteomyelitis: A rare complication of pancreatitis and PPP-syndrome

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ABSTRACT

Pancreatic diseases can be accompanied by periarthritis with bone necrosis and panniculitis (PPP-syndrome). It is postulated that this is caused by systemic activity of pancreatic enzymes leading to microcirculatory disturbances and fat necrosis. The morbidity and mortality of the PPP-syndrome is high. Successful treatment of pancreatitis can lead to resolution of accompanying panniculitis and periarthritis without adverse sequelae, but weeks or months after pancreatitis, asymptomatic necrosis of the bone may become symptomatic by fracturing spontaneously. In this report, we also describe osteomyelitis as a severe septic complication of bone necrosis caused by pancreatitis, in one case as acute tissue necrosis and in another case months after pancreatitis spread haematogenously.

1. Introduction

Polyarthritis and/or intraosseous fat necrosis are very rare complications of pancreatitis [1]. If the therapy of pancreatitis is successful, most joint symptoms are transitory [2]. Osseous complications can occur weeks or months after pancreatitis, such as periosteal reaction, calcification of intramedullary lesions, joint space narrowing, metaphyseal collapse and spontaneous fractures. All of these are aseptic osseous lesions. Pancreatitis, however, is associated with a high incidence of sepsis and associated mortality and it is important to be alert to the possibility of secondary infection [3].

We therefore present cases of an early and a late haematogenous infection of pancreatitis-induced osteonecrosis and discuss both in light of current literature.

2. Case presentations

2.1. Case 1

A 52-year-old male patient presented with bilateral aching, swollen, erythematous wrists and ankles as well as red-nodular skin changes of the right distal lower leg. The lesions were increasing in severity and had started 5 days earlier. There was no abdominal pain or abdominal distension. Blood investigations showed high serum amylase [12.708 U/L; normal range (N) 30–118] and lipase (21.405 U/L; N 13–60) as well as total leukocyte count of 32,100 cells/μL (N 4300–10,000) and CRP 35 mg/dL (N < 0.5). The abdominal CT scan showed a florid exsudative pancreatitis with cysts in the pancreatic head (3.5 × 5 cm) and a distended pancreatic duct (5 mm). X-rays showed no changes of the extremities. Conservative treatment with volume resuscitation and antibiotics was started while the patient was febrile at 40 °C. A therapeutic trial of prednisolone 80 mg/d was commenced for 3 days. Stenting of the pancreatic duct was unsuccessful due to swelling of the ampulla of Vater. In the next 3 days, the wrists and ankles had swollen further to the point of perforation (Fig. 1A and B). The MRI of the right ankle showed a pathological bone marrow signal with pronounced oedema and reduced fat, ankle joint effusion, a peritendinitis of the peroneal tendons and diffuse swelling of periarticular soft tissue (Fig. 1C and D). On the fourth day of admission, we were forced to incise both wrists and ankles because of perforations and compartment syndrome. Intraoperative findings were of purulent liquid fat necrosis, necrotic periostium, necrotic tendon sheaths and joint capsules as well as fat-free white necrosis of cancellous bone. Microbiological investigations were sterile. Histological investigations showed typical fat necrosis in the soft tissues and in cancellous bone (Fig. 2A and B). During treatment, the osseous necrosis became infected and showed the histologic pattern of purulent destructive osteomyelitis.

Under conservative treatment of pancreatitis (volume management, antibiotics with meropenem and vancomycin), amylase and lipase returned to normal and acute pancreatitis was healed within 2 months. CT-morphologically the cyst in the pancreatic head had now grown to 7 cm. As the proximal pancreatic duct had widened, another ERCP was performed which showed several scarred stenosis of the distal pancreatic duct. Stenting was still not possible. As pancreatic enzymes were normalizing, a resection of the pancreas

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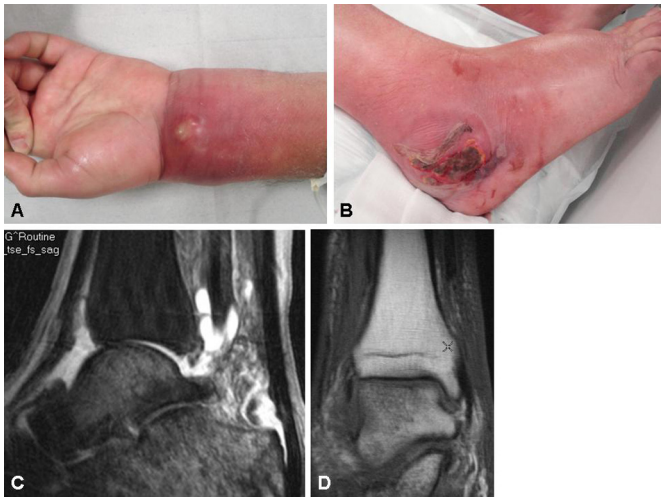

Fig. 1. Clinical findings 3 days after admission of the patient (case 1). Massive swelling on the left wrist (A) and on the right ankle with perforation of soft tissue necrosis (B). MRI of the right ankle shows in the T2-fat-suppressed weighting (C) in talus and calcaneus a pathologic cancellous bone signal with extended edema and reduced fat. In addition, the T1-weighting (D) shows an inhomogenous attenuation of the signal in talus and calcaneus with suppression of fat-mark.

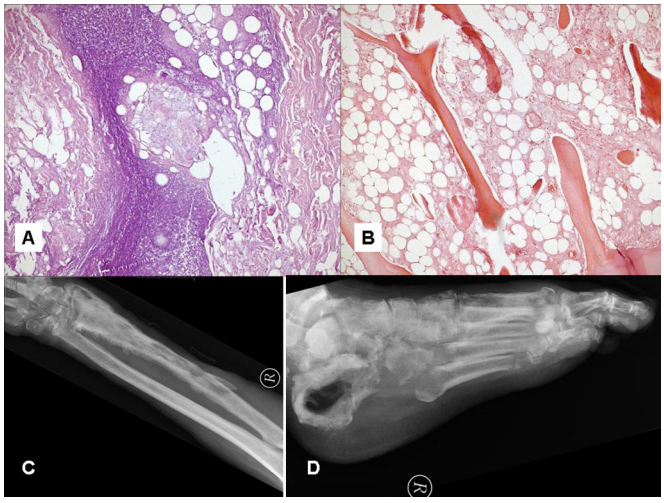

Fig. 2. Light micrograph of tryptical fat tissue necroses in soft tissue of the right lower leg (A) and in the right calcaneus (B) (hematoxylin–eosin stain, × 50). Conventional radiologic state of the patient 3 months after start of treatment (case 1). Right wrist and forearm a.p. with demineralisations of the wrist-near bones and a spontaneous fracture of the right distal radius (C). Right foot oblique with demineralisation and defect-osteolyses of the tarsal bones (D).

or punction of the cyst was not performed. The upper extremities could be conserved with little functional limitation by secondary healing and split skin grafting. Demineralization of wrist bones and a spontaneous fracture of the right distal radius were seen radiologically (Fig. 2C). In the lower extremities, infectious intraosseous fat necrosis and osteonecrosis developed which resulted in enzymatic destruction of joint capsules and caused joint empyemas (Fig. 2D). After 4 and 6 months, lower leg amputations were necessary. After 7 months, the patient was discharged into rehabilitation, having normal pancreatic enzyme activity but being left with bilateral lower leg prostheses.

2.2. Case 2

A 60-year-old male patient was admitted to hospital with abdominal pain. The abdominal CT scan showed an exudative pancreatitis thought to be caused by hyperlipidemia [triglycerides 5219 mg/dL (N < 200), cholesterol 1232 mg/dL (N < 200)] and/or alcohol abuse (alcohol delirium). Pancreatic enzymes could not be measured due to the massive hyperlipidemia in the acute phase of pancreatitis. Signs of panniculitis and periarthritis were not clinically apparent. Pancreatitis was treated conservatively by plasmapheresis, volume resuscitation and antibiotics. Two months later, the same patient was readmitted with symptoms of sepsis. *Staphylococcus aureus* was grown from blood cultures. The source was most likely a chemotherapy access port, which had been implanted 5 years earlier for colon carcinoma. After removal of the port, the symptoms of sepsis subsided. A control CT scan of the abdomen showed pseudocysts in the pancreas.

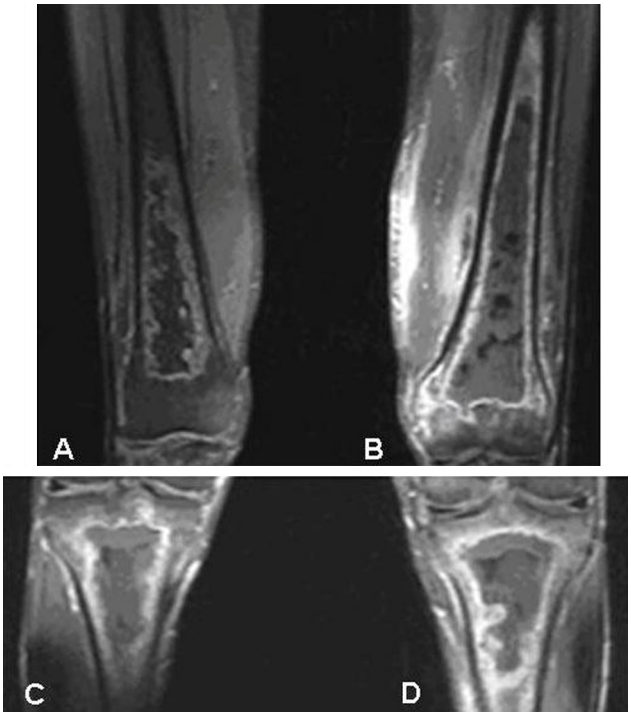

Fig. 3. MRI of the right knee (A, C) and left knee (B, D) (case 2). Liquid-filled hypointense content of osseous necroses with marginal contrast enhancement indicating osteitis. Central accumulation with peripheric contrast enhancement in the left musculus vastus medialis indicating a soft tissue abszess (B).

Six months after the subsiding of acute pancreatitis, the patient complained of knee pain, but no associated clinical or X-ray abnormalities were found. After 9 months, the patient developed an abscess of the soft tissues of the left lower leg, which was drained. As the abscess recurred 12 months later, an MRI of both lower extremities was performed which showed extended, liquid-filled osteonecrosis in both knee joint metaphysis (Fig. 3A–D). Aspirates of the osseous necrosis grew *S. aureus*. The antibiotic resistance profile matched the *S. aureus*, which was found in the blood cultures 2 months after the pancreatitis subsided. The infected osteonecrotic areas were debrided and laid with gentamycin beads. Until the infection had abated, the healing of the wound was delayed. Histology revealed a purulent osteomyelitis.

3. Discussion

The complete triad of the PPP-syndrome occurs very rarely together [4]. Until now, only 30 case reports have been published [5–9]. The disease can appear at any stage of life although it appears more often in the fifth decade of life. Most often men with chronic alcohol-induced pancreatitis are affected [10]. About two third of the patients have absent or mild abdominal symptoms [2]. The high