CASE REPORT

RJME
Romanian Journal of Morphology & Embryology
http://www.rjme.ro/

Frostbite arthropathy – a rare case of osteoarthritis, review of the literature and case presentation

LÁSZLÓ IRSAY[1,2], RODICA ANA UNGUR[1,2], ILEANA MONICA BORDA[1,2], ALINA DENIZA CIUBEAN[1], IULIA MOLDOVAN[2], MAGDALENA RODICA TRĂISTARU[3], KAMAL CONSTANTIN KAMAL[4], DIANA KAMAL[5], VIORELA MIHAELA CIORTEA[1,2]

[1]*Department of Physical and Rehabilitation Medicine, "Iuliu Haţieganu" University of Medicine and Pharmacy, Cluj-Napoca, Romania*

[2]*Department of Physical and Rehabilitation Medicine, Rehabilitation Clinic, Cluj-Napoca, Romania*

[3]*Department of Physical and Rehabilitation Medicine, University of Medicine and Pharmacy of Craiova, Romania*

[4]*Department of Family Medicine, University of Medicine and Pharmacy of Craiova, Romania*

[5]*Department of Physical and Rehabilitation Medicine, "Sama" Clinic, Craiova, Romania*

Abstract

Frostbite affects more commonly the northern population then it was suspected earlier, but wherever cold winter occurs, cold caused lesions are reported. Most often, it is described as soft tissue lesions, but deeper structures like tendons, ligaments, muscles, cartilage or bones can be affected. All extremities can be involved; lesions can lead to necrosis and amputations. First documented cases were described during military actions, but occupational or recreational activities can also be a risk factor for frostbite. Frozen or frostbite arthropathy is a rare cause of osteoarthritis. Usually, arthritis appears after a long time after frostbite, it can be decades apart. Frostbite arthropathy can result in different debilitating conditions. The current review describes the most important changes in frostbite and a rare but very serious late complication, which lead to arthropathy.

Keywords: frostbite arthropathy, osteoarthritis, disability.

Introduction

Prolonged exposure to low temperatures leads variably to skin lesions, but also to lesions of the deeper, subcutaneous, musculoskeletal structures. Joint and bone lesions appear late after months or years after the exposure in more than half of the patients [1].

The initial phases of frostbite can be categorized according to four degrees of severity:

(*1*) Cooling and freezing – vasoconstriction and vasospasm followed by transient arteriovenous shunting. If freezing persists, this phenomenon loses effect and the temperature in the tissues drops below -2ºC, and crystals are formed. Intracellular crystals destroy cell membranes, and extracellular ones, produce stasis with intracellular dehydration by osmotic mechanism. Symptoms include numbness and paresthesia. Pain appears after rewarming.

(*2*) The process of heating the limbs begins by absorbing heat. The intracellular and extracellular crystals melt, and the extracellular edema appears concomitantly with increased capillary permeability. Extravasations of fluids results in vesiculation, edema and loss of several layers of skin.

(*3*) Loss of entire skin thickness, with icy white digits. Tissue lesions appear in cascade through inflammation, stasis, thrombosis and necrosis. Decreases the level of prostaglandin E2 (vasodilator and antiplatelet effects), increases the level of prostaglandin F2α and thromboxane B2 (vasoconstrictor and platelet-aggregating effects).

(*4*) The consequences of cellular death appear. Immediate bone and cartilage involvement, with necrosis and subsequent loss of the affected part [2, 3].

Clinical changes in frostbite arthropathy are similar to those seen in hand osteoarthritis. In young people, the premature closure of the growth cartilages and secondary brachydactyly are described. Radiological changes are more frequent distally, asymmetrical and less affect the thumbs, probably due to palm protection [4, 5].

The treatment of frostbite arthropathy includes non-pharmacological (physiotherapy and surgery) and pharmacological treatment. There are no guidelines in this regard. Often non-steroidal anti-inflammatory drugs (NSAIDs) remain ineffective. A case has been described that responded to the administration of bisphosphonate, respectively Clodronic acid [6].

Case presentation

The 35-year-old male patient, known with proximal phalanx amputation of the second left finger after grade III degeneration with gangrene (2002), prosthesis of the proximal interphalangeal (PIP) joint of the left 4th finger (silicone prosthesis – February 2019), was admitted in our Department for mechanical pain and functional impotence in both hands, associated with the progressive swelling of the 2nd–5th PIP joints started in 2012.

We excluded inflammatory and metabolic causes of arthritis.

Physical examination of the hands revealed: spontaneous bilateral pain, which aggravated on palpation of metacarpophalangeal (MCP) joints, proximal or distal interphalangeal (DIP) joints, squeeze test was negative, swelling of PIP joints 2nd–5th fingers bilaterally, Heberden & Bouchard arthritic nodules, proximal phalangeal amputation of 2nd left finger (Figure 1). The examination of other joints was unremarkable.

Goniometer values on admission and upon discharge after 10 days of treatment are shown in the table below (Table 1).

Table 1 – ***Goniometer measures for hands joints, on admission and discharge***

Hands joints	Admission	Discharge
Left hand		
2nd MCP	90°	90°
3rd MCP	90°	90°
PIP	40°	55°
DIP	60°	60°
4th MCP	90°	90°
PIP	60°	80°
DIP	55°	55°
5th MCP	90°	90°
PIP	70°	80°
DIP	55°	70°
Right hand		
2nd MCP	90°	90°
PIP	40°	40°
DIP	65°	65°
3rd MCP	90°	90°
PIP	50°	50°
DIP	90°	90°
4th MCP	90°	90°
PIP	90°	90°
DIP	80°	90°
5th MCP	90°	90°
PIP	90°	90°
DIP	90°	90°

MCP: Metacarpophalangeal; PIP: Proximal interphalangeal; DIP: Distal interphalangeal.

The Disabilities of the Arm, Shoulder and Hand Score (DASH) score on admission 40; upon discharge 29 (after 10 days of treatment).

Hands radiographs in antero-posterior view, before the arthroplasty of the 4th finger, described the amputation of the proximal phalanx of the left 2nd finger, advanced arthritic changes at the level of the bilateral PIP joints. We did not consider the radiological reassessment important from the point of view of diagnosis and prognosis (Figure 2).

The histopathological exam, which was performed after the arthroplasty, revealed several changes in the cartilage, subchondral bone and other articular structures. The biological material, consisting of bone and cartilage fragments, containing both the necrosis-affected tissue and adjacent areas, was fixed in 10% neutral buffered formalin, fractionated, decalcified using trichloroacetic acid in increasingly higher concentration, included in paraffin, and after that, cut into 4–5 μm sections. The slides were then stained with Hematoxylin–Eosin (HE) and Goldner–Szekely (GS) trichrome, in order to visualize the microscopic aspects of the damaged area and to assess the changes, which appear in the adjacent areas.

The histological changes have varied from one area to another. There were still some areas where the cartilage was less affected by the pathological process. At this level, we noticed chondrocytes in chondroplasts, with a predominantly linear arrangement; however, even in this tissue we observed a reduction of the basophilia around the chondrocyte globules, which means a reduction of the territorial area and an increase of the conjunctival matrix at the level of the inter-chondroplastic area. All these aspects denote an incipient cellular suffering (Figure 3).

Near the affected tissue, the chondroplastic territorial area becomes smaller, the inter-territorial area has a non-homogeneous appearance, the chondrocytes have deformed nuclei with smaller dimensions than normal (Figures 4–6).

Appearance from the cartilage area where massive tissue disorganization was observed, showed missing isogenic series, fundamental substance with inhomogeneous aspect, cells with reduced appearance, disorganized, necrotic, anucleate, or pyknotic nuclei. Other aspects found at this level were marked reduction of cellularity, the remaining cellular elements had a modified structure, numerous anucleate cells, large areas with predominantly fibrillar structure (Figures 7 and 8).

However, in the cartilaginous tissue next to the subchondral bone, we noticed conjunctive-vascular buds that penetrate the cartilage, from the underlying bone, with the purpose of reshaping and consolidate the affected area (Figures 9 and 10).

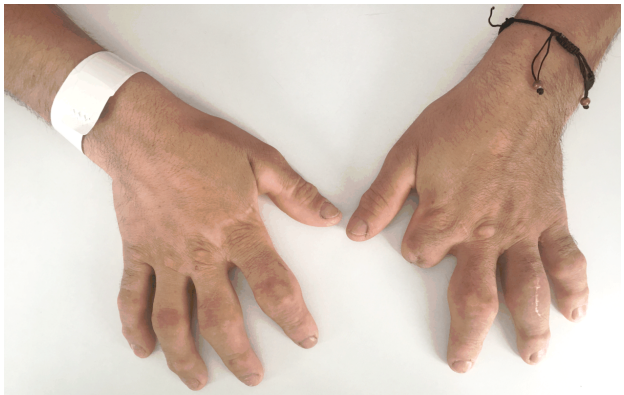

Figure 1 – ***The predominant swelling of the PIP joints, hindering the full extension of the fingers. PIP: Proximal interphalangeal.***

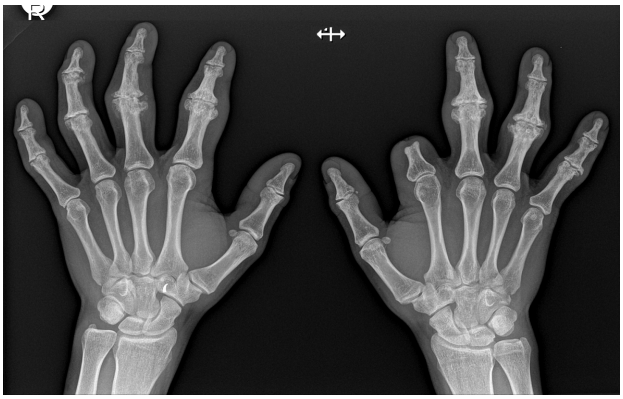

Figure 2 – ***Antero-posterior hands X-ray, before arthroplasty, showing amputation of the proximal phalanx of the left 2nd finger and advanced arthritic changes in bilateral PIP and DIP joints. PIP: Proximal interphalangeal; DIP: Distal interphalangeal.***

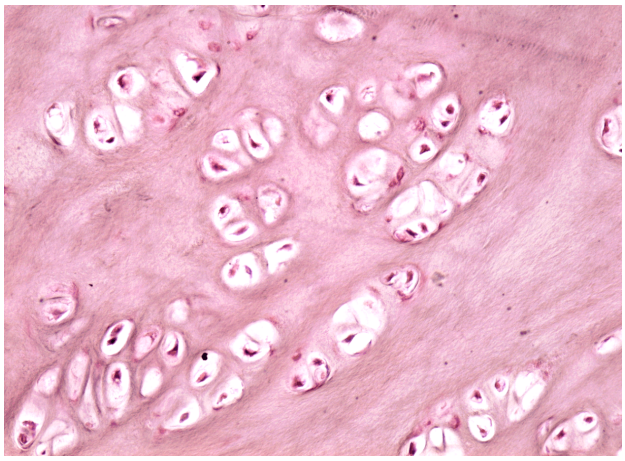

Figure 3 – *Cartilage area less affected by the pathological process. Chondrocytes in chondroplasts, with a predominantly linear arrangement, are highlighted. A reduction of the basophilia around the chondrocyte globules can be observed (HE staining, ×200).*

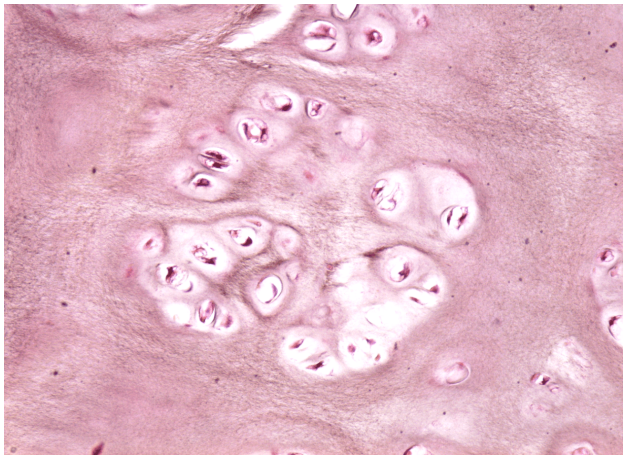

Figure 4 – *In this area of the cartilaginous tissue, the chondroplastic territorial area is smaller, the interterritorial area has a non-homogeneous appearance, the chondrocytes have nuclei with smaller dimensions than normal, pyknotic, deformed (HE staining, ×200).*

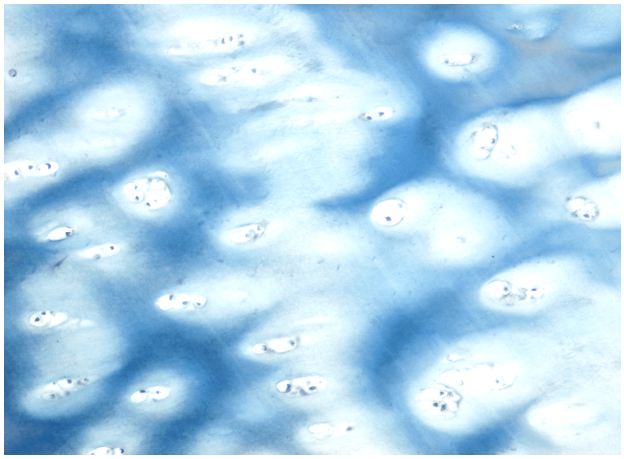

Figure 5 – *Appearance from the area adjacent to the necrotic process, with inhomogeneous arrangement of the fundamental substance, small cells with hyperchromic nuclei, sometimes anucleate (GS trichrome staining, ×200).*

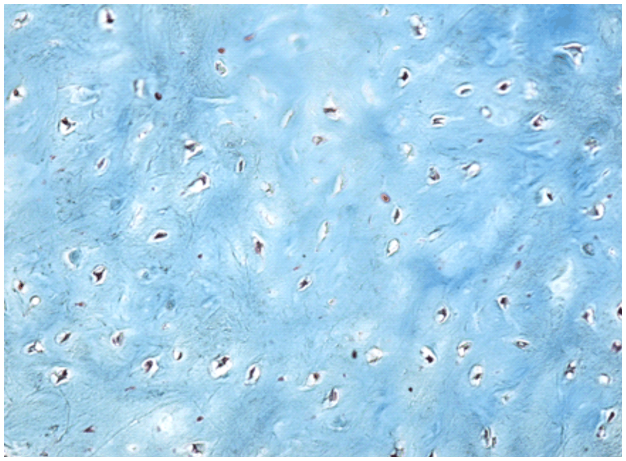

Figure 6 – *Area of cartilaginous tissue in which the overall suffering can be observed, the fundamental substance with inhomogeneous aspect, cells with reduced cytoplasm, pyknotic nuclei (GS trichrome staining, ×200).*

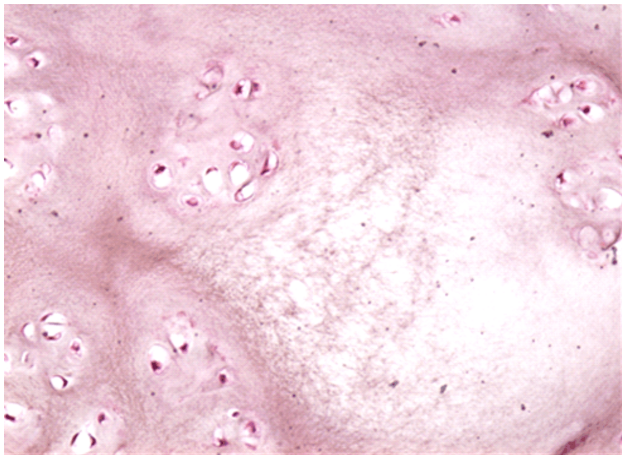

Figure 7 – *Appearance from the area exposed to the necrotic process. There is an intense area with predominantly fibrillar structure, a diminished number of chondrocytes, with modified structure, pyknotic nuclei, hyperchromic. Due to the marked destruction of cellularity and structural changes, no fundamental substance is produced anymore (HE staining, ×200).*

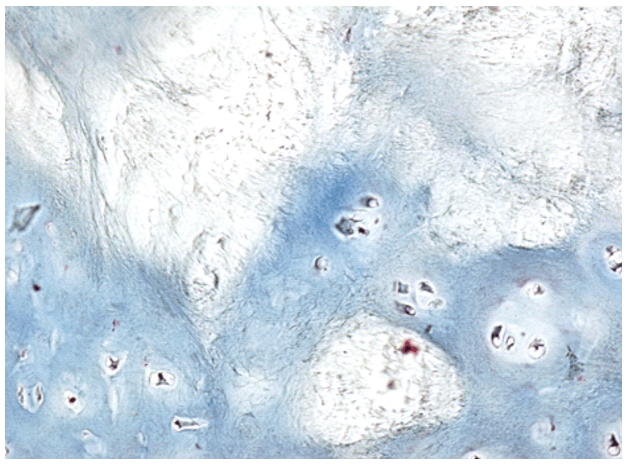

Figure 8 – *Aspect from the cartilage affected by the pathological process, marked destruction of the tissue, respectively of the fundamental substance, rare conjunctive cells, predominantly fibrillar tissue area can be noticed (GS trichrome staining, ×200).*

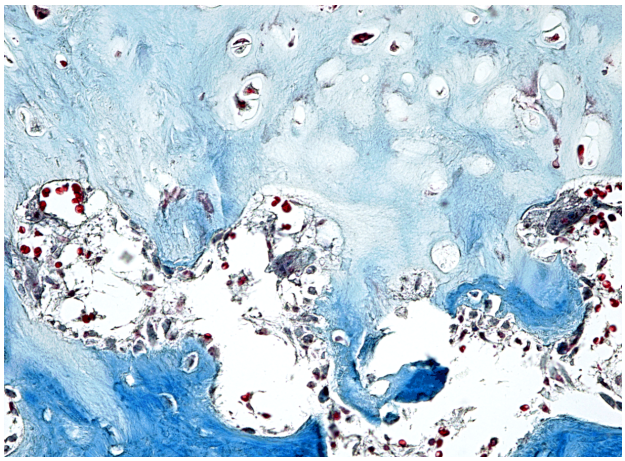

Figure 9 – ***Cartilage remodeling area. In the disorganized acellular area of the cartilaginous tissue, from the underlying bone tissue, vascular buds penetrate this area, attempting to remodel and consolidate the affected tissue (GS trichrome staining, ×200).***

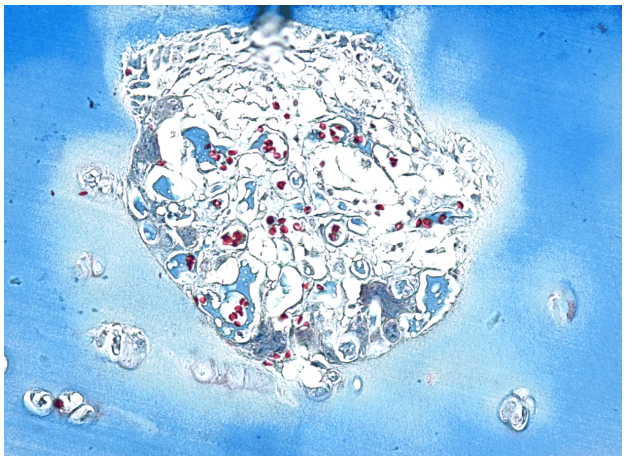

Figure 10 – ***Detail of the affected area of the cartilaginous tissue, of a conjunctive-vascular bud, which comes from the subiacent bone tissue in order to stop the destructive process. Red blood cells can be seen in the lumen of the vessels inside the conjunctival-vascular bud (GS trichrome staining, ×200).***

Due to the incomplete finger mobility, ultrasound evaluation was hampered. However, the examination showed a narrow joint space at the PIP joint level, with irregular surfaces, important osteophytes on both bone slopes; the capsule was shifted by the articular panus, without intra-articular fluid in pathological quantity (Figure 11). Flexor and extensor tendons were dislocated due to bone production in multiple phalanges. No Doppler signal was noticed at the level of the interphalangeal joints, in both hands. At the left PIP joint 4th level, ultrasound image of total arthroplasty was seen (Figure 12).

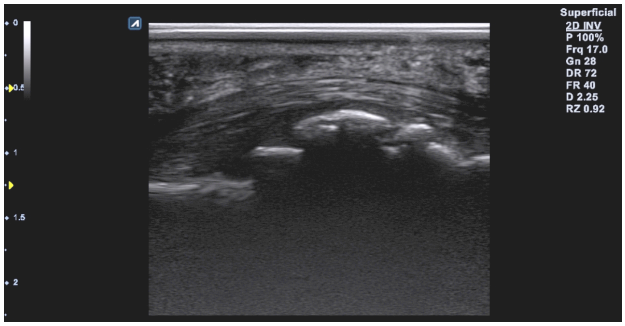

Figure 11 – ***Ultrasound image, with 18 MHz on right 3rd PIP joint, longitudinal section, narrow joint space, with irregular surfaces, important osteophytes on both bone slopes. PIP: Proximal interphalangeal.***

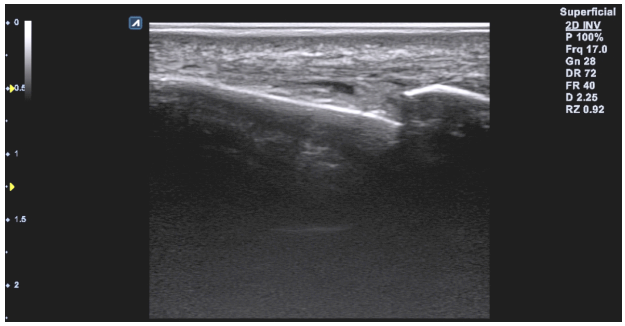

Figure 12 – ***Ultrasound image, with 18 MHz of PIP joint arthroplasty, longitudinal section, flexor and extensor tendons dislocated. PIP: Proximal interphalangeal.***

By excluding other common arthropathy and with positive history for frostbite lesions, the diagnosis was frostbite arthropathy. A physiotherapy program followed with the main objectives: to reduce pain and increase functionality. After 10 days of treatment, the evolution was favorable objectified by the goniometry and the DASH score. The long-term prognosis is reserved due to the slow progressive evolution of the disease that gradually leads to the loss of functionality.

Discussions

Hannibal lost half of his army crossing the Alps in 218 BC due to cold and frostbite secondary injuries. Napoleon's troops, Alexander the Great, or the American and British troops during the first and Second World War suffered from frostbite injuries [7].

Two US military doctors described bone damage after frostbite in 100 soldiers, during the Korean War between 1950–1951 [8].

One of the first documented frostbite arthropathy was perhaps of Pope Pius II who was forced to walk through snow close to Edinburgh during a visit in Scotland after the ship he was sailing, docked some 20 miles far from Edinburgh [9].

However, there are only a few case studies described in the literature and no well conducted trials [6, 10, 11].

Cumulative lifetime incidence of all types of frostbite may be as high as 44–68%.

Exposed anatomical structures to cold are at the highest risk of frostbite: the risk of finger frostbite increases linearly from 5% to 95% when skin surface temperature decreases from -4.8ºC to -7.8ºC. In case of frostbite, 1ºC difference between the cutis and subcutis can cause lesions.

Risk of frostbite is minor if the temperature is above -10ºC even with high airspeed [12].

Risk is higher in males, between 30–50 years, alcohol and drug abuse, mental illness, previous peripheral vascular disease, smoking (reduces nitric oxide level, which is a

vasodilator and increase the risk of thrombosis), race (African descent), contact with frozen materials (metals, water, ice, snow). Nonetheless, no clear methods are available to identify individual risk factors for vulnerability to cold injuries. A Norwegian study found an interesting aspect regarding smokers and smokeless tobacco (snus) – they are in the group of people with fast rewarm times after cold exposure. Another study (Miland & Mercer) presented that a window of at least four hours without smoking can lead to a speedier recovery in the elderly [13, 14].

Small bones of the hand and epiphyses, in children, are at high risk of frostbite. Prolonged exposure to cold can cause premature fusion and abnormal growth of the epiphyseal cartilage. Clinically is manifest as dwarfing and deformity of the affected limb and early onset arthritis [15].

Frostbite pathogenesis is described as a primary consequence of vasoconstriction that occurs after prolonged exposure to cold. Reheating leads to vasodilation with increased vascular permeability, fluid transudation and perivascular edema. Vascular stasis occurs with the accumulation of red blood cells and the appearance of fibrin deposits. Angiographic evidence of arterial spasm in the fingers and absence of blood circulation in the finger pulp was demonstrated [16].

Bone and joint changes appear as a consequence of vascular insufficiency with tissue anoxia, but also direct lesions of chondrocytes and their necrosis have been described. It is likely that this combination of vascular manifestations and tissue lesions results in changes in frostbite arthropathy, otherwise more patients with Raynaud's syndrome would have arthritis [4].

The earliest changes are soft tissue edema and loss of tissue. Osteopenia occurs relatively quick, then osteolysis and periostitis – these lesions appear approximately eight months after the exposure. In 7% of cases, there are subcutaneous gas bubbles due to bacterial infection, which is a bad prognosis factor and usually leads to amputations. Subsequent radiological changes are much like those we find in osteoarthritis of the hand, but much more pronounced and without preserving the symmetrical aspects. More specific and earlier modifications have been described using three-phase bone scintigraphy with Technetium-99m Methylene Diphosphonate (^{99m}Tc-MDP). An indicator of definitive healing is normal/high uptake in the phalanges. Second day examination can conclude weather amputation is needed. Efficacy of the treatment can usually be assessed between day 2 and 8 (some authors suggest 7) [6, 11, 17, 18].

Conclusions

Frozen or frostbite arthropathy is a rare cause of osteoarthritis, usually appearing a long time, sometimes decades after the initial injury. In order to correctly diagnose frostbite arthropathy in patients with finger or phalanx amputation, with history of frostbite, it is necessary to correlate the clinical, paraclinical, imaging and histopathological results.

Conflict of interests

The authors declare that there is no conflict of interests regarding the publication of this paper.

References

[1] Tishler JM. The soft-tissue and bone changes in frostbite injuries. Radiology, 1972, 102(3):511–513.
[2] McKendry RJ. Frostbite arthritis. Can Med Assoc J, 1981, 125(10):1128–1130.
[3] Lorentzen AK, Penninga L. Frostbite – a case series from Arctic Greenland. Wilderness Environ Med, 2018, 29(3):392–400.
[4] Pettit MT, Finger DR. Frostbite arthropathy. J Clin Rheumatol, 1998, 4(6):316–318.
[5] Wang Y, Saad E, Bonife T, Wainapel SF. Frostbite arthritis. Am J Phys Med Rehabil, 2016, 95(2):e28.
[6] Kahn JE, Lidove O, Laredo JD, Blétry O. Frostbite arthritis. Ann Rheum Dis, 2005, 64(6):966–967.
[7] Jarrett F. Frostbite: current concepts of pathogenesis and treatment. Rev Surg, 1974, 31(2):71–74.
[8] Vinson HA, Schatzki R. Roentgenologic bone changes encountered in frostbite, Korea 1950–51. Radiology, 1954, 63(5):685–695.
[9] Buchanan WW. Frostbite arthritis consequence of first papal visit to Scotland? Clin Rheumatol, 1987, 6(3):436–438.
[10] Turner M, Smith RW. Unusual and memorable. Erosive nodal osteoarthritis after frostbite. Ann Rheum Dis, 1998, 57(5): 271.
[11] Garg I, Baladron Zanetti MJ, Kendi AT. Bone scan in evaluation of bone viability in severe frostbite of the hand. Indian J Nucl Med, 2017, 32(4):367–368.
[12] Hassi J, Mäkinen TM. Frostbite: occurrence, risk factors and consequences. Int J Circumpolar Health, 2000, 59(2):92–98.
[13] Norheim AJ, Borud E, Wilsgaard T, De Weerd L, Mercer JB. Variability in peripheral rewarming after cold stress among 255 healthy Norwegian army conscripts assessed by dynamic infrared thermography. Int J Circumpolar Health, 2018, 77(1): 1536250.
[14] Miland AO, Mercer JB. Effect of a short period of abstinence from smoking on rewarming patterns of the hands following local cooling. Eur J Appl Physiol, 2006, 98(2):161–168.
[15] Crouch C, Smith WL. Long term sequelae of frostbite. Pediatr Radiol, 1990, 20(5):365–366.
[16] Gralino BJ, Porter JM, Rosch J. Angiography in the diagnosis and therapy of frostbite. Radiology, 1976, 119(2):301–305.
[17] Cauchy E, Chetaille E, Lefevre M, Kerelou E, Marsigny B. The role of bone scanning in severe frostbite of the extremities: a retrospective study of 88 cases. Eur J Nucl Med, 2000, 27(5):497–502.
[18] Cauchy E, Marsigny B, Allamel G, Verhellen R, Chetaille E. The value of technetium 99 scintigraphy in the prognosis of amputation in severe frostbite injuries of the extremities: a retrospective study of 92 severe frostbite injuries. J Hand Surg Am, 2000, 25(5):969–978.

Corresponding author
Alina Deniza Ciubean, Assistant Lecturer, MD, PhD Student, Department of Physical and Rehabilitation Medicine, "Iuliu Hatieganu" University of Medicine and Pharmacy, 8 Victor Babeş Street, 400012 Cluj-Napoca, Romania; Phone +40264–207 021, e-mail: alina_med87@yahoo.com

Received: November 4, 2019 *Accepted: February 16, 2020*