

Case Report

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Pathomorphological changes in the tibial metaphysis caused by a 40 years' osteoplastic material persistence after giant-cell tumor resection. Case report

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The authors describe a rare case of bone pathology in the tibial metaphysis bone tissue in a 75-year old female patient who underwent surgical treatment for a giant-cell tumor with further osteoplasty about 40 years ago. The biopsy material from the pathological focus was mainly represented by connective tissue. Cartilaginous and bone structures were not numerous. Pathological mineralization was a typical histological manifestation of degenerative changes in the pathological focus. There were no signs of the giant-cell tumor recurrence as well as no signs of inflammatory process. Persistence of unconsolidated osteoplastic material for a long time was the cause of severe right-side gonarthrosis that required knee arthroplasty.

Keywords Gonarthrosis, pathohistology, pathological mineralization

Osteoarthritis is a heterogenous group of diseases with multifactorial pathogenesis. It is characterized by destabilization of normal relations between the processes of degradation and synthesis of chondrocytes, and between the extracellular matrix of the articular cartilage and subchondral bone [3]. Among all the chronic diseases of the articular structures of the lower limb, osteoarthritis deformans of the knee or gonarthrosis occurs most frequently. The pathology may result from any changes in this articulation and is characterized by continuous progression [4]. The risk of osteoarthritis after treatment of giant-cell tumor around the knee is high. Giant-cell tumors mostly develop in humans who are older than 20 years. Limb salvage surgeries (bone marginal and intratumoral resections) are considered to be the main method of treatment in case of a benign process. Currently, a big choice of plastic materials is available to fill in the cavities to reconstruct the bone after tumor resection [2].

We present a clinical example of a rare case of bone tissue pathology revealed after knee joint arthroplasty.

Female patient U., 75 years old, was admitted to FSBI RISC RTO with complaints of pain in the knees, more severe on the right side, limitation of joint movements, and joint deformity. Pain was permanent but aggravated by weight-bearing. According to the patient's medical history, she underwent a surgical intervention for a giant-cell tumor in the upper third of the right tibia and further osteoplasty about 40 years ago when she was 30 years old.

Conservative treatment for pain had a transient effect. It appeared to be ineffective in 2015 when pain intensified.

Varus deformity of the right knee without swelling and a 1-cm shortening of the right lower limb was diagnosed during clinical examination. Perifocal tissues were moderately painful when palpated. The skin of the affected limb in the knee area had no scars, fistulas, and was of normal color. There were no vascular or

neurological disorders. The patient walked limping but without additional supports. Right knee range of motion was between 170 and 90°.

Gonarthrosis of grade III, joint gap narrowing, subchondral sclerosis, marginal bone overgrowth, the decrease in bone mineral density were identified in the X-rays (**Fig. 1**). The established diagnosis was: other primary gonarthrosis, grade III right side gonarthrosis, varus deformity, contracture of the right knee, and painful joint.



Fig. 1 X-rays of the right knee of a female patient U: before treatment, AP (a) and lateral (b) views

The X-rays showed the presence of a pathological focus in the right tibial metaphysis that was presented by dense and intensively mineralized fragmented masses in the focus center and a radiolucent zone along their periphery that reached the subchondral bone. No close connection of the mentioned masses with the surrounding bone was revealed.

Knee arthroplasty was performed in order to improve the functional condition and relieve pain. The surgery was cemented arthroplasty of the right knee using a posteriorly stabilized Zimmer NexGen LPS-Flex implant. The following components were placed: tibial component No 5, the right femoral component, 14-mm E insert, F insert No 5-6. The pathological focus in the bone metaphysis was removed using a curettage technique. The postoperative material was sent to a morphology laboratory for pathohistological investigation. The studies were performed in compliance with the Ethical Principles for Medical Research Involving Human Subjects, and in accordance with The Rules of Clinical Practice in the Russian Federation.

The harvested material was fibro-osseous friable fragments of different sizes (up to 2 cm in length) and of irregular shape. The material was fixed in a 10 % solution of neutral formalin, decalcified in the mixture of hydrochloric and formic acids, dehydrated in alcohols of ascending concentrations (from 70° to the absolute), impregnated with a sealing mixture and embedded in paraffin. Histological

sections from 5 to 7 µm thick were prepared using the sledge-type microtome of Reichard Company (Germany). The sections were stained with hematoxylin and eosin, and according to the Masson trichromic technique. The study and photomicrography of histological preparations were performed using the Opton photomicroscope (Germany), DiaMorph hard- and software complex (Moscow, Russia), and AxioScope.A1 stereo microscope supplied with an AxioCam digital camera and Zenblue software (Carl Zeiss MicroImaging GmbH, Germany).

The study of the material from the pathological focus revealed the areas of tissue-atypical mineralization located between loose gentle fibrous paucicellular connective tissue masses (**Fig. 2**) that stained in soft lilac color with hematoxylin and eosin, and light blue color when the Masson trichromic technique was used.

It should be noted that pathological mineralization was observed in the soft tissue and in the cartilaginous and bone structures as well. Small numbers of mineralized non-functional vascular structures were also identified in the areas of connective tissue mineralization (**Fig. 3**).

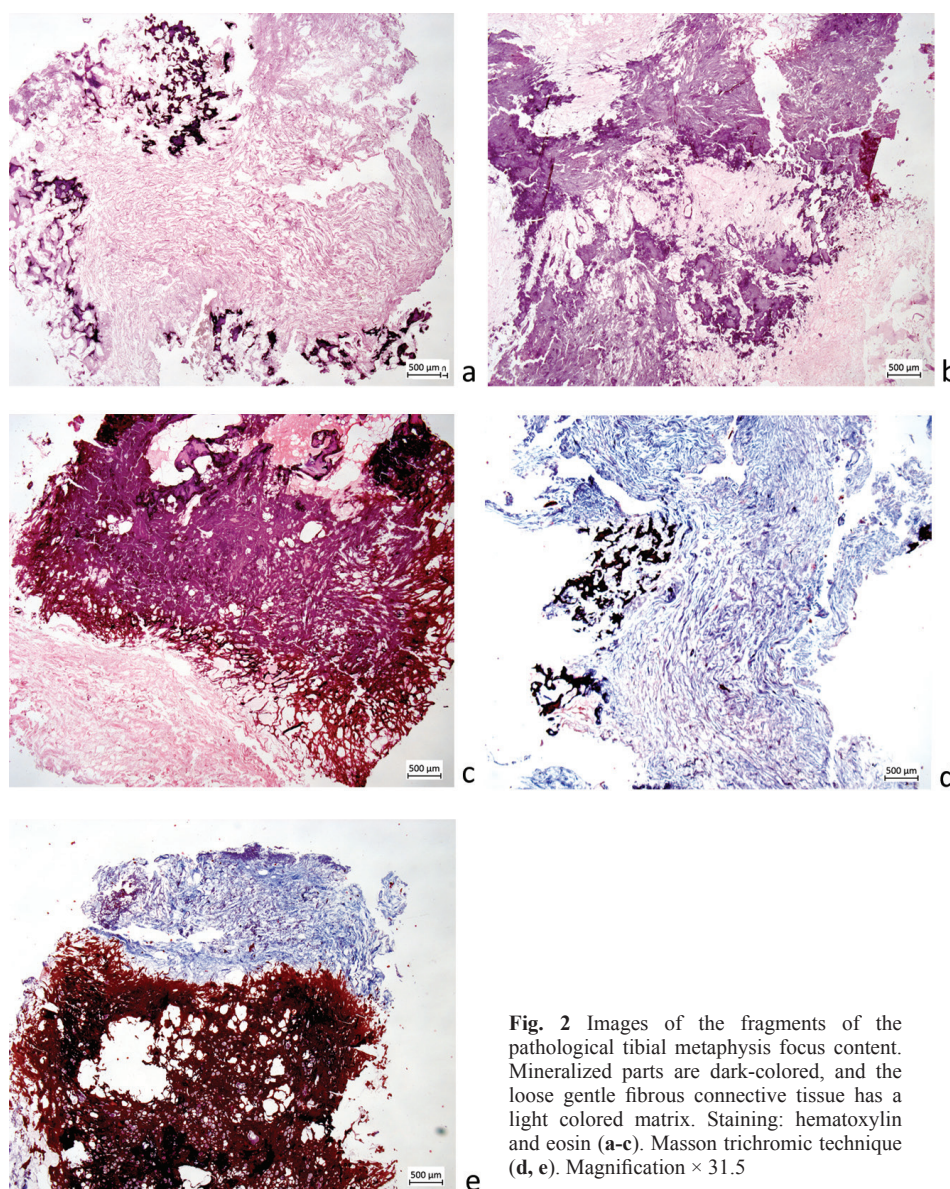


Fig. 2 Images of the fragments of the pathological tibial metaphysis focus content. Mineralized parts are dark-colored, and the loose gentle fibrous connective tissue has a light colored matrix. Staining: hematoxylin and eosin (**a-c**). Masson trichromic technique (**d, e**). Magnification × 31.5

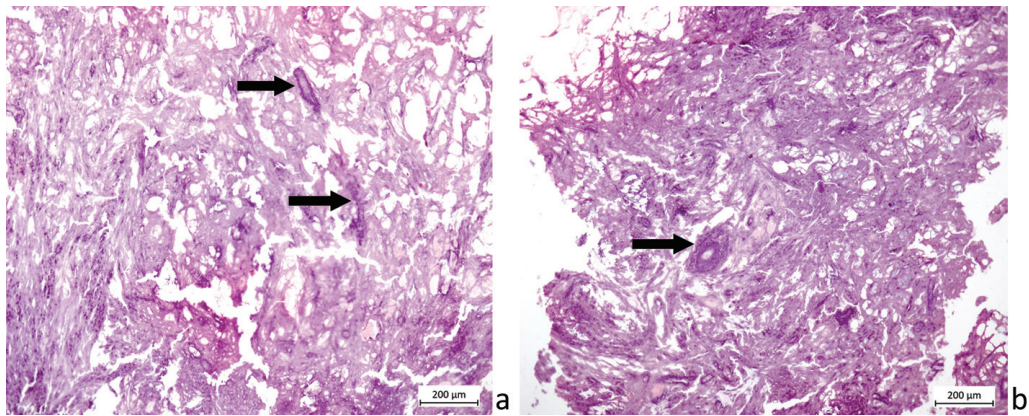


Fig. 3 Mineralized vascular structures in the connective tissue matrix undergoing mineralization (arrows). Staining: hematoxylin and eosin. Magnification $\times 125$

Mineralizing bundle structures were observed in the gentle connective tissue (**Fig. 4**) with some bundles oriented in the opposite direction to each other.

Less marked mineralization of the fibrous matrix was observed in some fields of its rarefaction. However, bundle organization was not formed in the fibrous part of the matrix but the structure acquired cellular organization (**Fig. 5**). Rounded cavitory structures of different sizes were located in the cellules of the net.

Chondroid structures were represented by fields of atypical tissue, bizarre in their configuration, with uneven contours and different matrix density (**Fig. 6**).

Moreover, the cartilaginous fields did not form a

typical hyaline structure and differed in varying colors characteristics of staining.

Bone structures were also characterized by atypical structure and a non-functional kind of matrix (**Fig. 7**). Moreover, bone structures were not numerous in the material studied.

Thus, the biopsy material from the pathological tibial focus was mainly a connective tissue of gentle fibrous type with few cartilaginous and bone structures. Their pathological mineralization was a typical manifestation of degenerative changes in the pathological focus. No signs of bone giant-cell tumor recurrence were found. Inflammatory process was not observed.

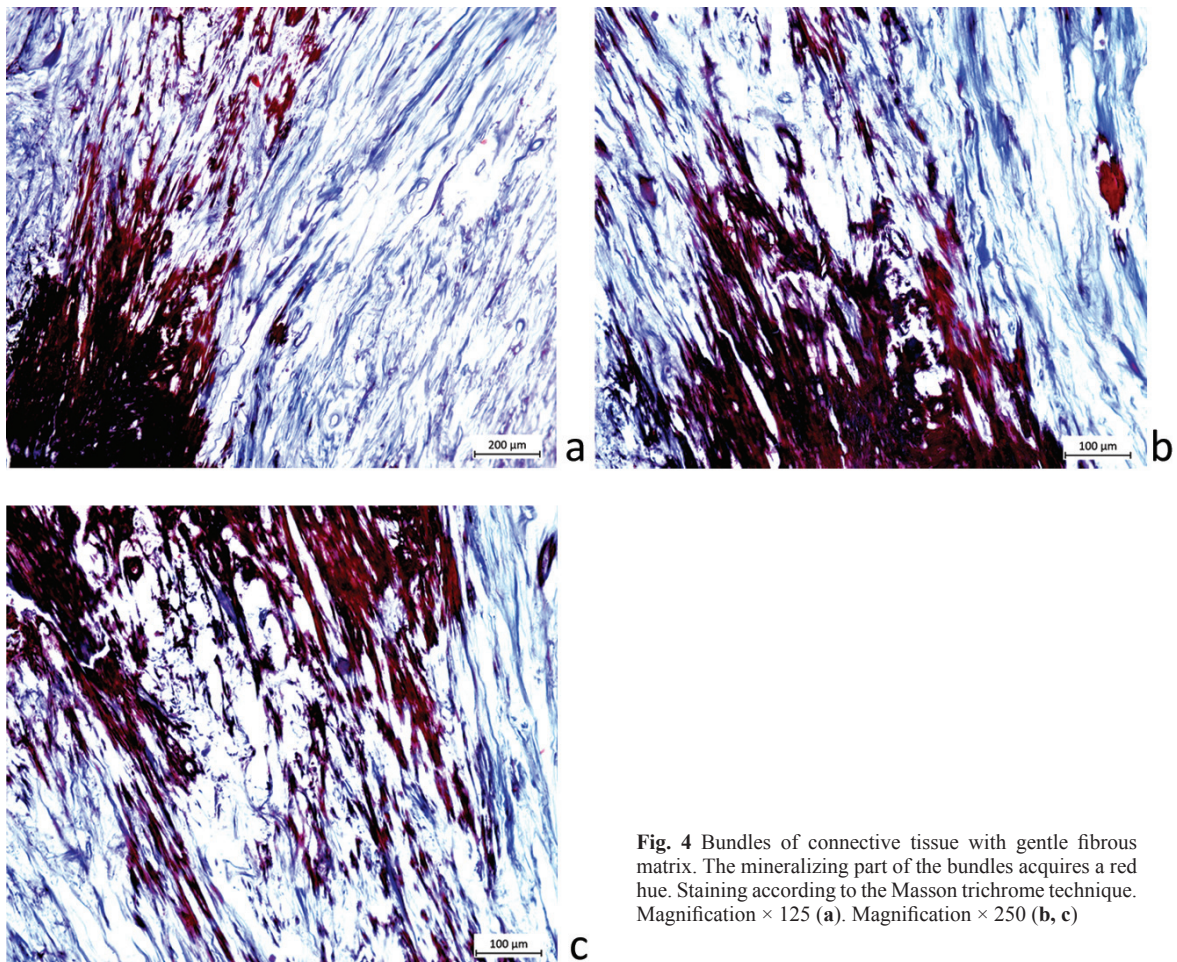


Fig. 4 Bundles of connective tissue with gentle fibrous matrix. The mineralizing part of the bundles acquires a red hue. Staining according to the Masson trichrome technique. Magnification $\times 125$ (a). Magnification $\times 250$ (b, c)

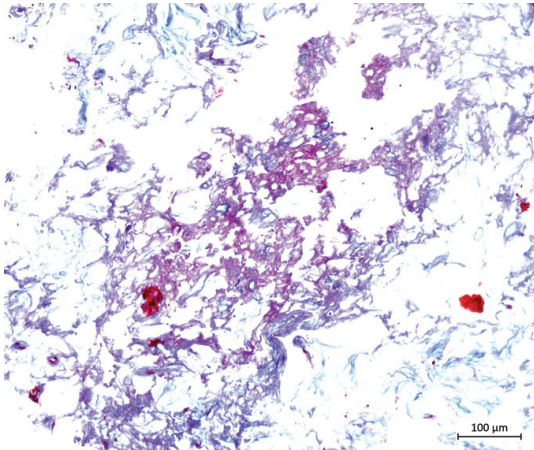
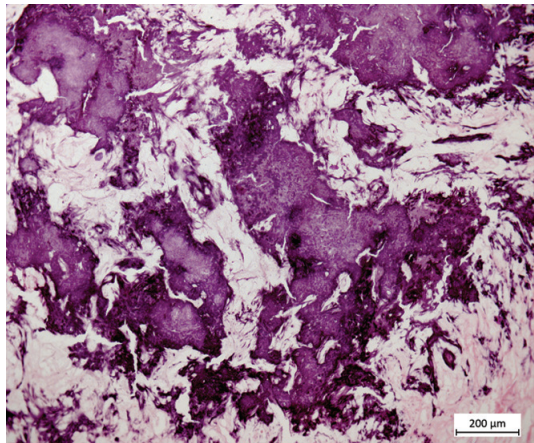
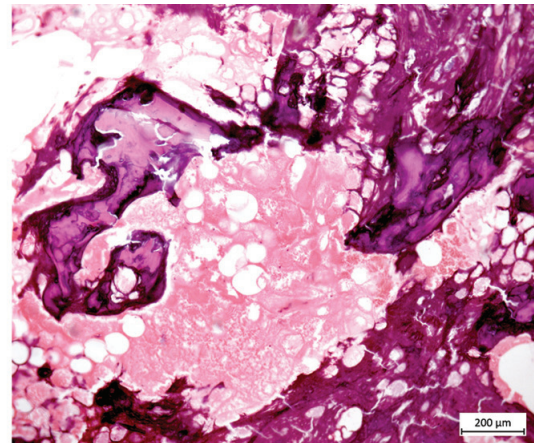


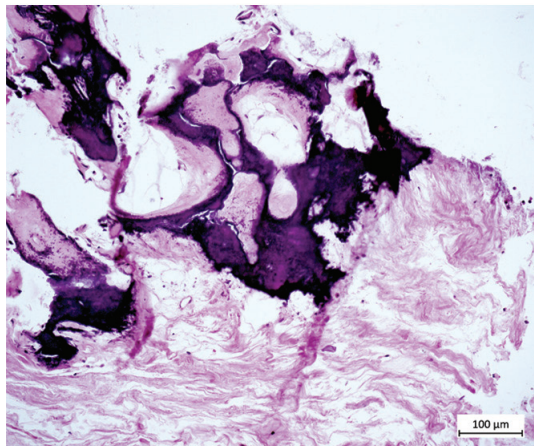
Fig. 5 Loose cellular organization of the collagen matrix with poorly marked mineralization (in the middle) as a field of reddish-lilac color. There are empty cavitory structures of rounded shape in the net cellules . Staining according to the Masson trichromic technique. Magnification $\times 250$.shape



a

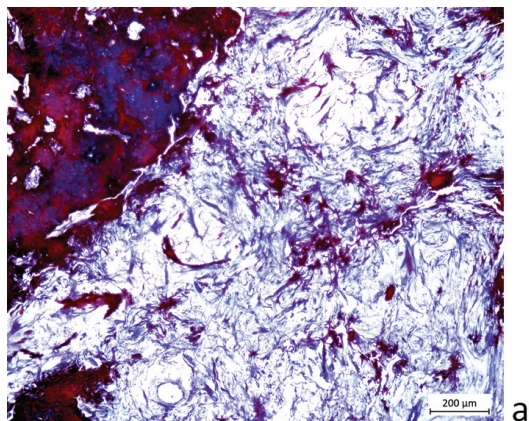


b

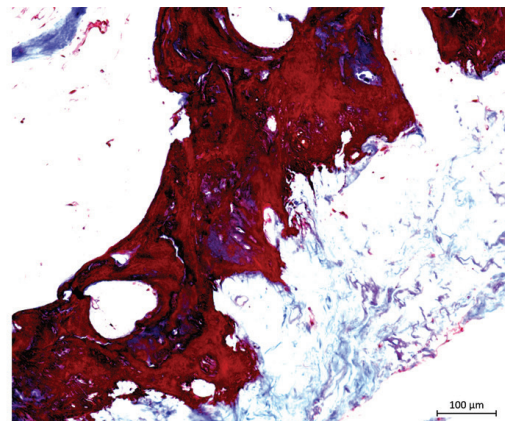


c

Fig. 6 Tissue-atypical cartilaginous fields with different mineralization density: Islets of cartilaginous tissue are not of a distinct hyaline phenotype (a). Cartilaginous structures with ossification areas, bone structures of a "blue" bone type. Staining with hematoxylin and eosin (b, c). Magnification $\times 125$



a



b

Fig. 7 Non-functional architectonics of bone structures. Staining according to the Masson trichromic technique. Magnification $\times 250$ (a). Magnification $\times 125$ (b)

In our opinion, the identified changes in the metaphyseal tibial bone tissue were the result of the previous surgical treatment (about 40 years ago) of a giant-cell tumor and osteoplasty after it. However, the type of the material used for osteoplasty remained unknown. According to the patient, the autologous graft was not harvested. It seemed that the unknown osteoplastic material did not have any osteoreparative impact on the focus of the earlier performed surgical intervention. Its prolonged persistence was accompanied by gradually developed dystrophic process with pathological mineralization and slow lytic effect on the surrounding bone tissue followed by changes in the adjacent tissues and destructive influence on the structural and functional status of the knee joint that resulted in gonarthrosis.

We think that the morphological characteristics of the pathomorphological changes that were revealed are similar to the changes in a so-called Nora disease (Nora's lesion. Bizarre parosteal osteochondromatous proliferation – BPOP) [5]. The similarity lies in the morphological manifestations only while clinical data and location are different from our case. Thus, in the cases of Nora's lesion, as a rule, the paraosseous structures of the hand and foot phalanges are involved; bone and cartilaginous structures grow exophytically and do not have any close contact with the adjacent bone [1, 5]. In the case presented by us, atypical tissue structures were located intraosseously but did not have any close contact with the surrounding bone.

CONCLUSION

Osteoplastic surgical interventions may not result in a reparative effect in some cases. Persistence of unconsolidated osteoplastic material for a long time that undergoes progressive dystrophic reactions might cause

destructive pathological processes in the surrounding bone structures. It was the reason of severe right side gonarthrosis that required knee arthroplasty in the case that has been presented.

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