INTRODUCTION
Carpal intraosseous ganglia (IOG) are rare pathological structures in the hand, which most commonly involve the lunate bone [1-3]. Several terms have been used in literature to describe the intraosseous ganglion cysts, including synovial bone cyst, juxta-articular bone cyst, ganglionic cystic defect of bone and subchondral bone cyst [4]. Their pathogenesis has not been fully elucidated, although several theories of their formation exist. It is thought that they are the result of synovial herniation, proliferation of synovial rest cells, metaplasia of precursor cells of mesenchymal origin, neoplasia or traumatic mucoid degeneration of connective tissue [5,6]. IOG contain viscous fluid rich in hyaluronic acid and have a thin wall or lining membrane comprised of fibroblasts and collagen fibers [1,5,6]. Normally, their clinical presentation involves chronic pain in the wrist, accompanied by radiological lucency in the carpal bones [7]. Very rarely, these anomalies can be present bilaterally [8]. The differential diagnosis includes simple bone cysts, post-traumatic cysts, giant cell tumors and enchondroma [9,10]. Conservative therapy is the first line of treatment for both asymptomatic and symptomatic lesions and includes non-steroidal anti-inflammatory drugs combined with periodical wrist immobilization [11]. If symptoms persist for more than 6 months despite the conservative treatment, surgical treatment is indicated. It consists of curettage of the cyst and application of a bone graft, most often an autologous graft is performed by an open surgical approach. Complications are rare and include joint stiffness and impairments of the vascular system of the lunate bone [12]. Another treatment option is an arthroscopic minimally invasive technique, which involves debridement and grafting of the lunate IOG [1].

CASE REPORTS
Case 1: A 31-year-old, right-handed woman presented with a 4-month history of activity-related pain, classified as moder-
ate (6 points according to the visual analogue scale, VAS) and pain under pressure in her right wrist over the lunate bone. She had no history of trauma and inflammatory disease. Physical examination revealed that both flexion and extension range of motion were limited (flexion 40°, extension 60°) in comparison to the left wrist (normal range of motion - flexion 80°, extension 85°). Pinch and grip strength in the affected hand were measured with a Jamar dynamometer and a decrease in the values was observed (18 kg for grip strength and 3 kg for pinch strength). Laboratory tests were within the normal ranges. These included routine examinations - complete blood count (CBC), biochemical tests and inflammatory markers (CRP, ESR and fibrinogen). X-ray of the hand did not reveal a pathologic lesion within the carpal bones (Fig 1a, b). However, magnetic resonance imaging (MRI) visualized the lesion within the lunate bone and the presence of fluid (Fig. 1c).

Figure 1: a) Antero-posterior radiograph; b) Lateral radiograph; c) MRI visualization of the lesion within the lunate bone.

Surgical treatment was performed through a dorsal approach. A fluid-filled soft tissue mass in the lunate bone was observed. Excision of the ganglion cyst and curettage of the affected bone were performed. The wound was closed in the usual manner. A short arm plaster cast was applied for three weeks. The postoperative period was uneventful. Physical therapy consisted of three sessions lasting for 10 days, conducted every three weeks. Each daily session included the following sequence of therapeutic procedures: 15-20 minutes of low-frequency impulse magnetic field, followed by 2 cycles of cryotherapy for 3 minutes each in order to alleviate pain and finally active kinesiotherapy against minimum resistance and with minimum range of motion lasting for approximately 25 minutes. The resistance was gradually increased over the next few days together with the range of motion and finally reached a maximum duration of 45 minutes. The patient was educated into everyday activities and underwent functional labour therapy. On the same upper limb, active exercises in full range of motion were conducted for the elbow and shoulder joint. Together with all these therapeutic procedures, active kinesiotherapy against resistance was performed on the contralateral upper limb. As a result of this complex treatment the range of motion was greatly improved (wrist flexion range of motion was up to 78° and extension range of motion went up to 85°), previous symptoms, such as pain and impaired function of the wrist were resolved and full grip and pinch strength was restored three months after operation (40 kg for grip strength and 6 kg for pinch strength). No clinical recurrence was detected on 9 months follow-up.

Case 2: A 26-year-old, right-handed girl presented with a 14-month history of right wrist pain, classified as low intensity pain (3 points according to VAS) and a radiolucent cystic lesion affecting the lunate bone. (Fig 2a, b) Right wrist flexion range of motion was measured as being 50° and extension range of motion 60°. Grip and pinch strength were measured with a Jamar dynamometer and were found to be slightly diminished (20 kg for grip strength and 3 kg for pinch strength).

Figure 2: a) Antero-posterior radiograph; b) Lateral radiograph.

Laboratory tests (CBC, biochemical tests, and inflammatory markers) were within the normal ranges. The preoperative diagnosis of an IOG affecting the lunate bone was accepted. The patient was operated through a vertical dorsal approach. A fluid-filled soft tissue mass in the lunate bone was detected. A typical gelatinous ganglion fluid was curetted and the lining membrane was removed. We also observed signs of aseptic inflammation with thickened synovial membrane. Afterwards, the capsule-ligament apparatus was repaired. A short arm plaster cast was applied for three weeks, as in the previous case. The postoperative period went smoothly. Physical therapy started after 21 days of immobilization and was conducted along the described pattern. The combination of surgical treatment and physical therapy yielded good results, with alleviation of pain and restoration of range of motion to normal values (flexion 85°, extension 90°). Grip and pinch strength were measured and showed a marked improvement (41 kg for grip strength and 6.5 kg for pinch strength). There were no complications or local recurrence on 8 months follow-up. In both cases the histopathological findings were consistent with an IOG and revealed a thin-walled cyst, filled with homogenous eosinophilic acellular gelatinous matter. The wall was built of compressed collagen fibers, fibroblasts and mesenchymal cells and had no true epithelial lining.

DISCUSSION

IOG of the carpal bones is a rare cause of chronic wrist pain [1,4,7]. Most commonly IOG affect the lunate, followed by the capitate, scaphoid, triquetrum and trapezoid bones [1-3,7,13].

The IOG of the carpal bones are classified into two types. According to one classification, in terms of histopathology, type I is a multiloculated cavity, surrounded by a fibrous membrane, which contains gelatious material; its wall is composed of connective tissue cells, similar to synovial cells, as well as inflammatory cells. Type II, on the other hand, is described as ‘bone cyst-like pathologic change’ and its center is composed of fibrous connective tissue instead of gelatinous material. The solid contents resemble the thick inner wall of juxta-articular bone cysts. Its internal structure is composed of dense fibrous tissue and collagen fibers [14]. Another classification also distinguishes between two types of IOG - one, which represents an intramedullary penetration of a soft tissue ganglion and is associated with bone erosions and radiographic lucency and another, which shows no signs of an extraosseous component and is thus termed as ‘idiopathic’ [8,15]. In line with these divisions, we classified the IOG reported herein as type I under the classification of Ikeda and Oka and as type II according to Pablos et al., since no extraosseous components of soft tissue ganglions were observed [8,14]. The most often encountered of these soft tissue ganglions is the dorsal ganglion, which usually originates in the scapholunate interosseous ligament. This offers an explanation as to why the lunate and scaphoid are the most commonly involved carpal bones [15].

The differential diagnosis of an IOG affecting the lunate bone is very broad as other processes can also present with chronic wrist pain, hyperaemia, swelling and functional impairment of movements in the wrist joint. Review of literature data supports the proposal of the following classification of the pathologic conditions, which are included in the differential diagnosis: a. benign bone tumours (chondroma, enchondroma, chondroblastoma, osteoid osteoma, osteoblastoma, giant cell tumor of bone, desmoid fibroma); b. non-neoplastic lesions (rheumatoid arthritis-associated conditions, osteoarthrosis-related conditions, osteomyelitis, pigmented villonodular synovitis, crystalline deposition disease); c. various cysts (juxta-articular bone cyst, unicameral or solitary bone cyst, aneurysmal bone cyst, necrobiosis pseudocyst); d. other diseases such as Kienböck's disease [12,14-16]. The correct diagnosis is established through several imaging modalities [12]. Conventional radiographs of IOG affecting the lunate bone present eccentrically located well-defined osteolytic lesion, outlined by a sclerotic rim [8]. Periosteal reaction, expansion of the cortex or internal calcification is not observed [16]. Sometimes, these ganglia can be occult on standard radiographs and are detected on CT and MRI only [8]. CT and MRI present additional imaging data which are vital for the accurate preoperative planning [16]. MRI images demonstrate intermediate signal intensity on T1-weighted images and high signal intensity on T2-weighted images [8]. Bone scintigraphy can also provide additional data supplementing the diagnostic process. A marked focal increase of the tracer uptake is observed with this imaging study [15].

Surgical treatment of the IOG of the lunate bone is indicated even in asymptomatic cases if continuous monitoring of the patient through radiography reveals progressive growth of the cyst, because it leads to bone replacement and erosion, resulting in pathologic fractures [15,16]. Classical surgical treatment of lunate bone lesions presenting with chronic wrist pain includes open curettage and bone grafting [1,12-15]. It is often done by autologous bone grafts, which offer excellent biological and mechanical properties but are associated with higher risk for donor site morbidity and limited material availability [17]. In comparison, calcium phosphate bone cement is a synthetic bone graft which is useful for repairing bone defects after curettage of an IOG or another type of bone cyst of the carpal bones [3]. Our experience and review of literature data show that traditional open surgery is a successful method with good postoperative results and low risk of recurrence, which allows a definitive histological diagnosis and is therefore often the predominant method of choice [1,11,12]. Its disadvantages include longer recovery period and a higher risk of joint stiffness due to the plaster cast immobilization. Arthroscopically assisted minimally invasive technique of debridement and grafting of the IOG has also been reported as a safe method with minimal morbidity, scar complications and recurrence risk, which reduces the operative area and minimizes post-operative joint stiffness by using a bandage rather than cast immobilization [1,12]. Moreover, this surgical technique allows a complete observation of the joint and in case of concomitant synovial cysts, these can be treated simultaneously [12]. Arthroscopic surgery however, does not permit the establishment of histological diagnosis [12]. In rare cases, dorsal flap arthroplasty, prosthetic replacement, radiocarpal or intercarpal fusion could be performed, as alternative treatment [2-4,7].

CONCLUSION

Intraosseous ganglia of the lunate bone are rarely encountered lesions which may be either asymptomatic or presenting with chronic wrist pain, swelling and functional impairments. They involve several histopathological types whose pathogenesis is not yet fully known. The differential diagnosis of IOG is complex and requires the use of numerous imaging modalities. Various surgical techniques, including open curettage and bone grafting or arthroscopically assisted surgery are used with low risk of recurrence. The presented case reports demonstrate the use of traditional open surgery with good
postoperative results and its advantages in establishing the definitive histological diagnosis. Knowledge of such lesions of the carpal bones has great clinical significance because it is associated with frequently encountered symptoms and its treatment most often leads to complete cure of the pathological condition.

REFERENCES