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Rehabilitation and clinical evolution aspects in a case of Osteoid Osteoma

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Abstract: Introduction. Osteoid osteoma represents about 3% of all primary bone tumors and 11% of all benign bone tumors. Data from the literature suggest that a neuromuscular rehabilitation program after osteoid osteoma surgery is very beneficial and improves the general quality of life**.**
Material and methods. A 30-year-old male patient with intermittent right shoulder pain radiating to the right hand, and recurrent myalgias in the past year presented to our neurological department. The neurological examination highlighted limitation of the abduction of the right upper limb. The paraclinical investigations included plain radiography of the right upper limb and electroneurography, which were normal, and native cervical MRI which revealed discrete C5 disc overflow, without visible signs of compression. The patient presented limited initial response to NSAIDs, so his treatment was changed to corticosteroid therapy. Further, the patient was guided to undergo a rheumatological examination where a musculoskeletal ultrasound was performed, showing no any specific modification. Additionally, we indicated a native right shoulder MRI, which revealed a signal modification of the proximal humeral diaphysis. We further indicated an MRI scan with contrast of the upper right limb, which revealed a nidus at the top one-third of the humerus. Additionally, a CT scan with contrast of the same region displayed images that were highly suggestive of osteoma. The patient was referred to the orthopedics department, where a complete resection of the tumor was performed, and the pathology report confirmed the final diagnosis of osteoid osteoma.
Conclusions. Recovery after osteoid osteoma surgery is more beneficial if the neuromuscular rehabilitation program, that has an important role in increasing muscle strength, is combined with orthopedic devices and pain medication.

Keywords: bone tumor, osteoid osteoma, upper limb, imaging changes, differential diagnosis,

Introduction

Osteoid osteoma is a benign bone tumor that consists of bone forming tumor cells that are engaged in osteoid and bone production giving rise to small 1.5-2 cm, well-demarcated lesions, with a predilection for long bones (1-3). It is typically found in children, adolescents, and young adults, with the age range between 10 and 35 years being more common in men. It is relatively frequent and represents about 3% of all primary bone tumors, and 11% of all benign bone tumors (2,4). According to the Musculoskeletal Tumor Society staging system for benign tumors, osteoid osteoma is a stage-2 lesion, being classified as cortical, cancellous, or subperiosteal, with cortical lesions being most common (5).

The main presenting complaint is pain, described as dull, unremitting, initially mild and intermittent pain that increases in intensity and persistence over time (6). It tends to become increasingly severe at night and is usually relieved by salicylates and nonsteroidal anti-inflammatory drugs (NSAIDs) (7). Computed tomography (CT) is the best imaging investigation for osteoid osteoma, as it clearly delineates the nidus (8).

Material and methods

Case presentation

A 30-year-old male patient, without any pathological history, whose occupations includes heavy physical activity, presented with intermittent pain in the right shoulder radiating to the hand, and recurrent myalgias, which started approximately one year ago. He had previously presented in another medical department, where he was diagnosed with gout, but the initial diagnosis was disproved by the plain radiography of the right upper limb, which did not highlight any modifications at that moment, and by normal blood tests. Because his symptoms persisted, he presented to our neurological department. The general examination did not reveal any pathological modifications and the neurological examination showed a limitation of the abduction of the right upper limb, without motor deficit or any changes in sensitivity. Blood analyses were within normal limits.

First, we performed a plain radiography of the right shoulder, which revealed mild shoulder osteoarthritis *(Fig.1),* and a plain radiography of the right upper limb, which did not reveal any alterations *(Fig.2).* Next, we performed a native cervical MRI that showed a discrete C5 disc overflow, without visible signs of compression and spinal static disorder (*Fig.3*). The electroneurography showed similar parameters in both upper limbs, and the motor and sensory nerve conduction showed a normal result. *(Fig.4).* In this context, we recommended NSAIDs. Under this treatment the patient's symptoms decreased, leading to a free-pain period. At the next presentation, the patient mainly complained of nocturnal pain, stabbing in character, of irregular duration from a few minutes to several hours, occasionally associated with reduced muscle strength, which was still relieved by NSAIDs. After three months, the patient presented a relapse of pain on abduction of the right upper limb, without motor deficit, but with associated vasomotor changes in painful episodes, like redness of hands and minimal edema associated with total remission when the pain episodes ended. Therefore, we considered changing the patient's treatment to corticosteroid therapy, which ameliorated patient symptomatology, but with limited effect over time. Further, we recommended a rheumatological examination and a musculoskeletal ultrasound, which did not reveal any additional information. Based on the patient's new symptomatology, his occupation, which included heavy physical work, the paraclinical evaluation and the persistence of the pain, we considered reflex sympathetic dystrophy as a possible differential diagnosis. We continued our investigations with a right shoulder native MRI which showed diffuse hypersignal with a stained appearance at the level of the proximal third of the humeral diaphysis (*Fig.6*). To obtain a better image of the lesion we recommended upper right limb MRI with contrast, which revealed diffuse bone marrow edema in the proximal third of the right humeral diaphysis, with about 50 mm below the surgical neck of a fusiform thickening of the antero-inferior cortex, at a distance of about 40 mm with a thickness of about 13 mm, with surrounding reactive sclerosis and a small central bone nidus (*Fig.7*). The radiologist recommended an additional CT scan with contrast of the upper right limb, which showed a thickening of the anterior cortex, at a distance of about 35 mm, a thickness of about 11 mm and a diameter of 15 mm, with the presence of a central bone nest of 5.5 mm, without obvious sclerotic reaction around the nest. Thus, the image was highly suggestive of osteoma *(Fig.8).*

Based on this diagnosis, we directed the patient to the orthopedics department for evaluation and surgical treatment. They opted for a complete resection, under general anesthesia, of the bone tumor located in the right humerus. The pathological result of the biopsy from the removed tumor showed anastomosed bone trabeculae, lined with osteoblasts arranged in a layer, and rare osteoclasts. The lesion presented large areas of

immature bone, and the stroma had areas with fibrosis and bleeding. The final diagnosis was osteoid osteoma (*Fig.9).*

The neuromuscular rehabilitation program

The objectives of the treatment were: pain reduction, increased joint amplitude, increased muscle strength, increased muscle tone, increased quality of life, and reintegration into the family and social environment. The physical therapy consisted of active-passive, active resistive, and active mobilizations of the upper right limb lasting for 30 minutes per session, and orthotic wearing for the prevention of articular stiffness.

The patient had a favorable postsurgical outcome. The symptoms resolved following physical therapy and treatment with analgesics, antibiotics and an anticoagulant. After two months, on post-op follow-up, the symptoms disappeared, the abduction of the upper limb was normal and painless, and the patient was able to return to work.

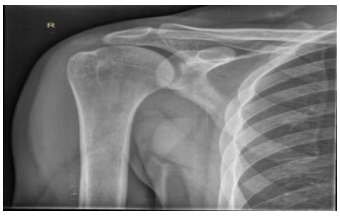

Fig.1. Plain radiography of the right shoulder – mild right shoulder osteoarthritis

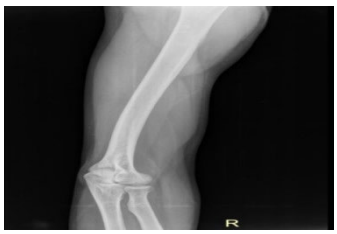

Fig.2. Plain radiography of the right humerus – normal aspect

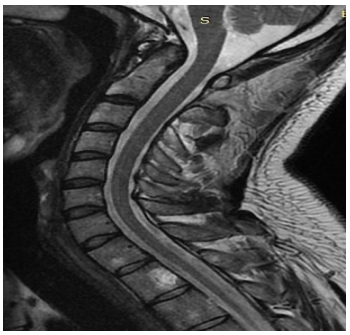

Fig.3. Native Cervical MRI - discrete C5 disc overflow, without visible signs of compression and spinal static disorder

Sensory Nerve Conduction:

Nerve and Site	Onset Latency	Peak Latency	Amplitude	Segment	Latency Difference	Distance	Conduction Velocity
Median.R							
Wrist	2.5 ms	3.3 ms	25 µV	Digit II (index finger)-Wrist	2.5 ms	140 mm	56 m/s
Ulnar.R							
Wrist	2.0 ms	2.7 ms	25 µV	Digit V (little finger)-Wrist	2.0 ms	115 mm	58 m/s
Radial.R							
Forearm	1.6 ms	1.6 ms	19 µV	Anatomical snuff box-Forearm	1.6 ms	100 mm	63 m/s
Lateral antebrachial cutaneous.R							
Elbow	1.9 ms	2.5 ms	13 µV	Forearm-Elbow	1.9 ms	120 mm	62 m/s
Median.L							
Wrist	2.8 ms	3.4 ms	21 µV	Digit II (index finger)-Wrist	2.8 ms	mm	m/s
Ulnar.L							
Wrist	2.0 ms	2.8 ms	15 µV	Digit V (little finger)-Wrist	2.0 ms	mm	m/s
Lateral antebrachial cutaneous.L							
Elbow	2.0 ms	2.5 ms	14 µV	Forearm-Elbow	2.0 ms	105 mm	53 m/s

Fig.4. Electroneurography - showing similar values for the right and left upper limb, in motor and sensory nerve conduction.

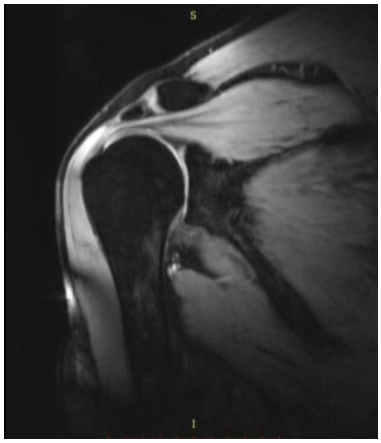

Fig.5. Native right shoulder MRI – diffuse hypersignal with a stained appearance in the proximal humeral diaphysis

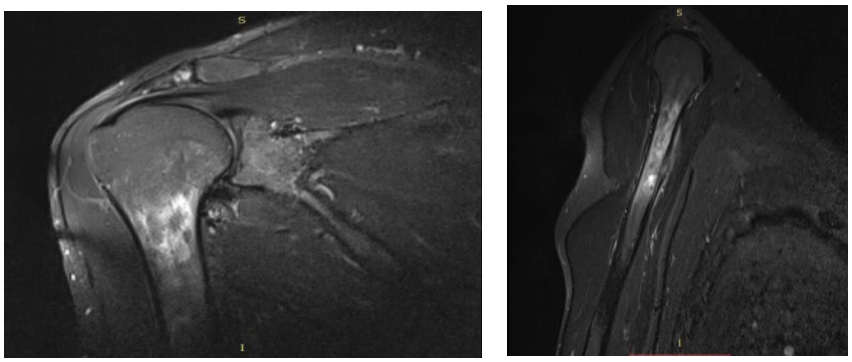

Fig.6. Right shoulder (left) and right upper limb (right) MRI with contrast – diffuse bone marrow edema in the proximal third of the right humeral diaphysis, fusiform thickening of the antero-inferior cortex, with surrounding reactive sclerosis and a small central bone nidus

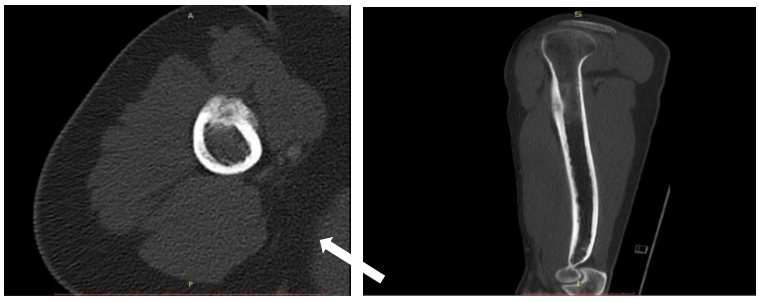

Fig.7. CT scan of the right upper limb with contrast – proximal third of the humeral diaphysis presents thickening of the anterior cortex; presence of a central bone nidus of 5.5 mm without obvious sclerotic reaction around the nest

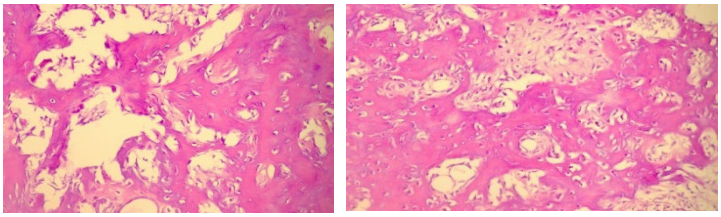

Fig.8. Hematoxylin-eosin stay, 20x, anastomosed and irregular osseous trabeculae with scattered osteoblasts and osteoclasts disposed in a fibrotic stroma

Discussion

The main differential diagnoses of an osteoid osteoma include chondroblastoma, bone infarction, Brodie's abscess, stress fracture, chronic osteomyelitis (9), but because of the lack of modifications in the initial paraclinical investigations, vague symptomatology, almost normal neurological examination and patient's occupation, none of these was taken into consideration in this particular case. The first diagnosis considered was cervical brachial neuralgia, due to the pain radiation and patient's occupation, but this was excluded by the native cervical MRI and the electroneurography, which did not reveal any radicular injury (10).

Then, due to the patient's persisting pain, accompanied by vasomotor changes, no nerve lesion observed in the electroneurography, and patient's occupation, which includes heavy physical work, the diagnosis of complex regional pain syndrome type I was considered, although the patient did not report having had any accident at work (11). Complex Regional Pain Syndrome is a neuropathic pain disorder defined by the presence of distinct clinical features including allodynia, hyperalgesia, sudomotor and vasomotor abnormalities, and trophic changes. There are two subtypes: type I, formerly known as reflex sympathetic dystrophy, and type II, formerly known as causalgia. Type I occurs in the absence of nerve trauma, while type II occurs in the setting of known nerve trauma (12). This diagnosis was excluded after the first MRI of the shoulder, which presented signal modifications.

The CT scan with contrast showed, at the level of the proximal third of the right humeral diaphysis, a thickening of the anterior cortex, at a distance of about 35 mm, a thickness of about 11 mm and a diameter of 15 mm, with the presence of a central bone nest of 5.5 mm, without obvious sclerotic reaction around the nest, which is highly suggestive of osteoid osteoma, without excluding the Brodie's abscess. Brodie's abscess is a sub-acute form of osteomyelitis, presenting as a collection of pus in the bone, often with an insidious onset (13). This diagnosis was excluded by the pathology result which highlighted small, circumscribed, anastomosing, irregular trabeculae of woven bone with variable mineralization, confirming the osteoid osteoma (14).

In osteoid osteoma, the pain usually occurs before the lesions are visible on radiographs. In this particular case, however, the time until the lesion was visible was of almost one year, which is an uncommon feature. Also, the lesion was not visible on a plain radiography, which is the initial imaging study of choice in this pathology (7,15,16). Usually, the pain is relieved by aspirin or nonsteroidal anti-inflammatory medication (NSAIDs), but in this case the response to NSAIDs was limited in time (17).

The treatment options include a non-operative approach with NSAIDs, which can accelerate spontaneous healing (18,19). However, in our patient's case, the effect of these drugs was limited in time. The surgical approach is an option for patients with severe pain and for those in whom the NSAIDs therapy failed. Available procedures include CT-guided radiofrequency ablation, *en bloc* resection, and CT guided percutaneous excision (20). *En bloc* excision of the tumor, cortical shaving and curettage of the nidus cavity are frequently used conventional techniques with positive outcomes. The tumor may be difficult to identify intraoperatively, and incomplete removal may result in recurrence (20,21). In order to reduce the surgical morbidity of open procedures, several percutaneous techniques using CT guidance have been used. The CT-guided Percutaneous Excision technique consists in a cannulated curette which is inserted into the lesion over a Kirschner wire under CT image guidance to excise the nidus (22). In CT-guided radiofrequency ablation, heat is applied locally to destroy the nidus. The radiofrequency probe is introduced into the nidus through a cannulated needle under CT-guided imaging, and the temperature at the tip of the probe is increased to approximately 90°C and maintained at that level for 5 to 6 minutes. The tip of the probe must be insulated to prevent injury to the soft tissues adjacent to the osteoid osteoma (23,24). In our specific case, the orthopedic team opted for *en bloc* resection, which lead to a complete resection of the tumor and a good outcome of the patient.

Conclusion

In conclusion, the complexity of the case was given by the non-specific symptomatology, the patient's occupation, which misguided the initial diagnosis towards a peripheral neuropathy, and the normal results of initial MRI scan investigations. The latency of almost one year from the initial symptoms to a visible lesion on the imaging examinations challenged us on the path to the final diagnosis. Also, the limited response to NSAIDs, and the long period from symptoms onset to diagnosis resulted in the patient's low quality of life. The rehabilitation program had an important role in increasing muscle strength and improving the general quality of life, through physical therapy, orthopedic devices and pain medication.

Informed consent - An informed consent was obtained from the patient participating in the study.

Declaration of conflict of interests The authors declare that there was no conflict of interest regarding the publication of this paper.

Author contributions.

1. Silvina Iluț: consultant neurologist in charge of the patient, conception and design of the case report. Approved the final version.
2. Gabriela Dogaru: drafted the discussion section and compared it to the current literature.
3. Oana Muresan: revised the case report.
4. Dafin Fior Mureșanu: coordinator of the team.

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