Bone Fractures in a 53-Year-Old Patient with Parathyroid Adenoma – A Case Report

Zdenko Ostojić¹, Maki Grle¹, Goran Moro¹, Zoran Zubak² and Marko Ostojić³

¹ Department of Orthopedics, University Hospital Mostar, Mostar, Bosnia and Herzegovina

² Special Hospital for Orthopedics, Biograd, Croatia

³ Faculty of Medicine, University of Mostar, Mostar, Bosnia and Herzegovina

ABSTRACT

The study reports a case of primary hyperparathyroidism in a middle-aged patient who was first admitted for persistent ankle pain and local swelling. The subsequent clinical procedures suggested cystic changes in several leg bones, which were later shown to be caused by the parathyroid adenoma. Clinical presentation of the primary hyperparathyroidism can be highly misleading, sometimes causing various clinical procedures before it is certainly diagnosed.

Key words: primary hyperparathyroidism, parathyroid gland, bone, Bosnia and Herzegovina

Introduction

The diagnosis of primary hyperparathyroidism is difficult to establish. This is reflected in the low number of published articles related to this subject, which is marked by the numerous difficulties. Most commonly changes associated with this condition are mistaken for tumor metastases, which can lead to various adverse outcomes. In the developed countries there is a number of possibilities to detect and diagnose such a disease, which is often detected in early stages without bone lesions. Developing countries do not employ screening on a regular basis, resulting in the later disease detection and usually more severe clinical manifestation of the disease¹. The aim of this study was to report a case, provide information on the diagnosis and treatment from the pathology and orthopedics point of view.

In 1981 von Recklinghausen described a bone disease and termed it *Osteitis fibrosa cystica*. In 1925 Mandl, surgeon from Vienna performed the first parathyroid exploration and removal of the parathyroid adenoma. He then noted improvement in the patient's bone status and managed to link the hyperparathyroidism to the bone disease. Later Albright described the classical clinical presentation of this entity in 17 cases. Today it is a disease that is often without any clinical symptoms and is rather rare in the developed countries. Various fractures as a consequence of the disease are rare, but if they do occur they most often affect spine.

Primary hyperparathyroidism is an endocrine disorder marked by the excessive parathyroid hormone secretion (PTH). The commonest causes of PTH secretion include adenomas (85%), hyperplasia (15%) or parathyroid carcinoma. The incidence of parathyroid adenomas is usually 42 cases per 100,000 persons, and it is three times more often in women. The main consequence of the PTH over-secretion is hypercalcaemia, which then affects several organ systems, including renal, skeletal, gastrointestinal and central nervous system. Following that, various clinical presentations may occur, including nephrolithiasis, diabetes insipidus, renal failure, osteitis fibrosa cystica, constipation, vomiting, nausea, lethargy, depression, delirium or coma. Primary hyperparathyroidism is most commonly detected in association with polyuria, polydipsia, muscular weakness, gastrointestinal problems, nephrolithiasis with laboratory tests showing increased levels of serum calcium. Yet, diffuse osteoporosis, cystic bone lesions and subperiosteal erosion are recorded in less than 2% of all cases².

Case Report

Patient, woman born in 1953, height: 169 cm and weight: 70 kg, reported to the clinic in February 2007, and stated that the reason for her admittance was persis-

tent pain in the right ankle, which has been present for few months. Physical examination revealed edematous region on the fibula several centimeters above the lateral malleolus, which was 2 by 3 cm, hard, immobile and diffuse. An X-ray revealed cystic changes of fibula and proximal part of tibia and also public bone, but without clinical symptoms suggesting the latter two. The combination of physical examination and radiographic findings suggested that the diagnosis was *Cystis fibulae lateri dextri/ Morbus Ollier*.

Patient was admitted later in April 2007, when a total body scan was performed. Main lab results were normal, with no other subjective disorders except the right ankle pain. However, before the body scan could be performed, the patient required surgical excision, which was performed in April in general anesthesia, with osteosynthesis with screws.

Next admittance was initiated in mid May 2007, when a pathology results came in, suggesting gigantocellular tumor, perhaps even brown tumor, but without a definitive diagnosis. The advancement of cystic changes caused another surgical treatment and reconstruction of the af-



Fig. 1. Ostheolithic lesions of the pelvic bone.



Fig. 2. Ostheolithic lesions of the distal part of fibula.

fected region of fibula (some 7–8 cm from malleolus). The affected region of the bone was supplemented with the graft from iliac bone, with the use of plates and screws. The conclusion of this procedure was that further tissue samples were needed, since the previous ones were not of sufficient quality for a definitive diagnosis to be set.

In June 2007 patient felt a strong pain underneath right knee. Physical examination revealed swollen right shin bone region, with crepitating. Radiological assessment revealed fracture without dislocation in the proximal part of the tibia where the previous x-rays suggested a large cystic lesion. After this, a radiologist suggested multiple fibrous affection of the skeleton, possibly even suggesting metastatic changes, the need to re-evaluate histological finding, serum calcium, PTH, and a thyroid scan.

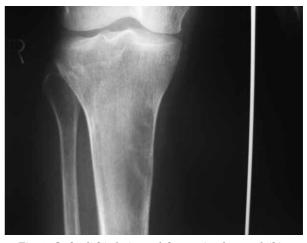


Fig. 3. Ostheolithic lesions of the proximal part of tibia.



Fig. 4. Proximal tibial fracture where lesion was previously described.



Fig. 5. Proximal tibial fracture where lesion was previously described.

Few days later, the patient was admitted for the third time, due to poor healing results of the tibial fracture. This resulted in the subsequent surgical approach, where two condilar plates were merged. Laboratory results suggested hypercalcaemia and elevated PTH level; calcium 3.82 mmol/L (lab range up to 2.6 mmol/L), PTH 751 pg/mL (lab range 10-69 pg/mL). Thyroid scan suggested adenoma of the lower right parathyroid gland. Following that, the histological results were available, suggesting gigantocellular tumor or an aneurismic cyst, albeit without a strong confirmation. In August, the patient underwent adenectomy of the right parathyroid gland, but the post-operative histological result suggested incomplete removal. The subsequent lab findings were in September PTH 738 pg/mL, Ca 3.35 mmol/L, P 0.69 mmol/L, while in October PTH was 1156 pg/mL, Ca $3.84~\mathrm{mmol/L},$ P0.67mmol/L (lab range 0.7-1.4 mmol/L; Table 1). Finally, in October the adenoma was removed using Gamma camera guidance.



Fig. 6. Regression of the ostheolithic changes of the pubic bone.



Fig. 7. Good treatment outcome of the fibula.

In December 2008 the patient reported a fall, leading to the severe trauma and right knee affection. The emergency admittance and knee punction was performed, without any positive finding. The leg was subsequently immobilized and the patient was discharged. Physical check-up examination suggested swollen knee with reduced movement. Radiogram suggested femur fracture without dislocation, which was surgically treated with screws. In November 2008 all laboratory findings were within laboratory range.

Discussion

Although primary hyperthyroidism is relatively well recognized clinical entity, it sometimes poses dilemmas even today. Two different types of bone lesions were described – slow and progressive³. Bone fractures are often recorded in the location of previous bone cysts. The developed countries have implemented screening calcium tests, thus reducing the possibility of severe complications, but in the developing countries disease may be di-

TABLE 1THE SEQUENCE OF THE LABORATORY FINDINGS IN THE IN-
VESTIGATED PATIENT

| Date | PTH | Ca | Р |
|-------------|------------|--------------|--------------|
| 20.11.2007 | 499 pg/mL | 1.82 mmol/L | 0.71 mmol/L |
| 28.12.2007 | 262 pg/mL | 2.19 mmol/L | 0.96 mmol/L |
| 26.02.2008 | 236 pg/mL | 2.23 mmol/L | 1.13 mmol/L |
| 22.04.2008 | 371 pg/mL | 2.23 mmol/L | 1.06 mmol/L |
| 27.05. 2008 | 249 pg/mL | 2.27 mmol/L | 1.05 mmol/L |
| 02.09.2008 | 231 pg/mL | 2.35 mmol/L | 0.80 mmol/L |
| | | | |



Fig. 8. Tibial fracture where the lesion was previously described, stabilized with two condilar plates. The lesion is gone, while the progression is satisfactory.

agnosed in an advanced state. Additionally, the diagnosis is usually set after half of year or so, which can lead to the number of various complications and fractures. This is why calcium should be included in the differential diagnosis of the ostheolithic changes, leading to the faster definitive diagnosis setting. Besides the primary hyper-

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In this case, the patient did not report any subjective discomfort. Histological finding suggested gigantocellular tumor, aneurismal cyst and also brown tumor. It becomes obvious that histological results may be misleading, especially in the cases when clinical, radiological or histological brown tumor can be the lead towards hyperparathyroidism⁵. In this case, the body scan suggested multiple fibrous dysplasia, although radiologist suggested that differential diagnosis could also include metastases, leading to the conclusion that the experience that the pathologist has can also facilitate the diagnosis.

Conclusion

Painful skeletal swellings that appear as osteolithic changes in the X-rays may suggest primary hyperparathyroidism. Further clinical protocol should include X--rays (pelvic, head, spine), biochemical tests (calcium, PTH), parathyroid ultrasound and scan. In the case when the diagnosis is confirmed, it is our opinion that osteolithic changes should be taken care of, preventing fractures. If these do appear, they should be treated with the AO methods. This is supported by the end result in the current study, with complete healing of the affected bones. We do hope that we will be able to include screening test and enable earlier diagnosis and reduce the number and the extent of complications which can arise as the consequence of the undiagnosed disease.

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Z. Ostojić

Department of Orthopedics, University Hospital Mostar, Bijeli brijeg bb, Mostar, Bosnia and Herzegovina e-mail: zdenkoostojic@net.hr

LOMOVI KOSTIJU KOD 53-GODIŠNJEG PACIJENTA SA PARATIROIDNIM ADENOMOM – PRIKAZ SLUČAJA

SAŽETAK

Ovaj rad prikazuje slučaj primarnog hiperparatiroidizma kod pacijent asrednje životne dobi koji je prvi put primljen u bolnicu zbog dugotrajnih bolova skočnog zgloba i lokalne otekline. Klinički postupci koji su uslijedili prikazali su cistične promjene u nekoliko kostiju nogu, za koje je kasnije pokazano da im je uzrok paratiroidni adenom. Klinička slika primarnog hiperparatiroidizma može biti maskirana, što može uzrokovati provedbu raznih kličinkih postupaka prije postavljanja definitivne dijagnoze.