

## Clinical case

## Pelvic chondroblastoma in an adolescent. New treatment approach

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**ABSTRACT.** Surgical management of tumors located in the spine and the pelvis involves greater difficulty. Moreover, these tumors are usually very large and vascularized. Preoperative embolization of the internal iliac artery is a relatively safe procedure that may reduce the risk of bleeding and local recurrence in the case of benign tumors. Chondroblastoma is a tumor that is rarely located in the pelvis; its more frequent location is the triradiate cartilage. We describe a case of a chondroblastoma with a relapsing aneurysmal cystic component in the acetabulum of an adolescent patient. Treatment consisted of embolization of the internal iliac artery, fluid hyperthermia, hydrogen peroxide and bone marrow application. The patient was found to be asymptomatic at the 5-year postoperative follow-up. The technetium ( $^{99m}\text{Tc}$ ) sestamibi scan was negative for tumor activity and found no lung metastases.

**Key words:** chondrosarcoma, pelvis, neoplasia, pain.

**RESUMEN.** Los tumores que están localizados en la columna vertebral y en la pelvis tienen asociada una mayor dificultad para el manejo quirúrgico. Además, estos tumores son habitualmente de gran tamaño y ricamente vascularizados. La embolización prequirúrgica arterial de la arteria ilíaca interna tiene la bondad de ser un procedimiento relativamente seguro que puede reducir el riesgo de sangrado y de recidiva local en tumores benignos. El condroblastoma es un tumor raro en la pelvis y la localización más frecuente es en el cartílago trirradiado. Se presenta un caso de condroblastoma con componente quístico aneurismático recidivante en el acetábulo de un paciente adolescente. Su tratamiento consistió en la embolización de la arteria ilíaca interna, hipertermia hídrica, peróxido de hidrógeno y aplicación de médula ósea. Con cinco años de evolución postoperatoria el paciente se encuentra asintomático. El rastreo con Sestamibi- $^{99m}\text{Tc}$  fue negativo para actividad tumoral y sin metástasis pulmonares.

**Palabras clave:** condrosarcoma, pelvis, neoplasia, dolor.

### Level of evidence: IV (Act Ortop Mex, 2011)

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### Introduction

The diagnosis of pelvic tumors may be delayed compared with long bone tumors because the clinical presentation is unusual in most cases and they may be mistaken for low back pain, arthritis, muscle contracture or alterations of the sacroiliac joint. External muscle tumors are not visible in the early stages and X-rays are often times misinterpreted as the osteolytic tumor lesion may be mistaken for intestinal air levels or may be hidden by the devices used to protect the ovaries or the testes. This may account for the high rate of soft tissue extension.<sup>1</sup>

Surgical management of the percentage of tumors located in the spine and the pelvis involves greater difficulty. Moreover, these tumors are usually very large and vascularized.

The spine and the pelvis are anatomical regions difficult to treat due to the need to protect the spinal cord, neurovascular elements and the immature osteocartilaginous structures, in the case of patients who are still growing.<sup>2</sup>

The nature of these tumors hinders hemostasis. Blood loss may reach volumes greater than 3,000 ml in 44.53% of patients in whom tumors in the acetabulum or the sacrum of up to 400 cm<sup>3</sup> are resected. Often times complete tumor resection is difficult, as well as the reconstruction of the bone defects, whether created or preexistent, involving surgeries lasting no less than 200 minutes.<sup>3</sup> The recurrence rate of lesions treated only surgically is quite high.

Due to the difficult surgical approach to these benign bone tumors, many surgeons have decided to treat them with radiotherapy. Complications and sequelae in immature skeletons are well documented (premature physeal closure, scoliosis, pelvic hypoplasia, coxa vara), as well as cystitis, proctitis and late sarcomatous degeneration.<sup>3</sup>

Arterial embolization to treat bone tumors was documented in 1975 by Hilal and Michelsen and then by Feldman, Casarella and Dick at Columbia-Presbyterian Medical Center. There is much more experience published about the use of arterial embolization for the treatment of gastrointestinal bleeding, refractory epistaxis, pelvic fractures and, more effectively, for arteriovenous malformations.<sup>2</sup>

It has been described that selective preoperative arterial embolization not only prevents bleeding, but it also plays a role as adjuvant in the treatment of large pelvic tumors.

Preoperative arterial embolization of the internal iliac artery has the advantage of being a relatively safe procedure that may reduce the risk of local relapse in cases of giant cell tumors and other benign tumors.<sup>4</sup>

According to Enneking and Dunham's classification (1978), modified by Sanjay et al. (1993), primary pelvic tumors are located and resected by regions. Region I is the sacroiliac area; region II, the periacetabular area, and region III, the ischiopubic area. The decision of selective arterial embolization is based on the proximity of tumors to the major iliac vessels as seen in MRI or CT angiography.

Excluding metastatic disease and myeloma, the most frequent primary malignant lesions in the pelvis, according to Unni (1996) and Campanacci (1999), are chondrosarcoma (28%), osteosarcoma (21%), and Ewing's sarcoma (18%). In two large series of benign bone tumors of the pelvis, 17% were located in the ilium, 14% in the sacrum, 4% in the pubic bone, and 3% in the ischium. Campanacci's series on benign tumors of the pelvis and hip reports that osteoid osteoma (33%) was the most frequent tumor, followed by osteochondroma (22%), giant cell tumor (19%), and chondroblastoma (7%). Among pseudotumor lesions, the most frequent one in patients with persistent growth cartilage in the ilium was Langerhans cell histiocytoma (eosinophilic granuloma), followed by the simple or aneurysmatic bone cyst.<sup>5</sup>

In patients with persistent growth cartilage the most frequent benign bone tumors are aneurysmatic bone cyst, os-

teoblastoma, giant cell tumor, and chondromyxoid fibroma; a large percentage of the latter are located in the spine and pelvis. The treatment of choice is local resection, curettage or bone graft, if necessary.<sup>2</sup>

Chondroblastoma is an infrequent tumor representing around 1% of benign bone lesions, but it may progress to malignant chondrosarcoma. Around 60% of cases occur during the first and second decades of life; it is more frequent among males. The lesion is found predominantly in the epiphysis or apophysis. Its incidence is high in the femoral head or the tip of the greater trochanter, the proximal humerus, and the femoral and tibial condyles, but it may also be found in other bones (skull, posterior vertebral structures and tarsal bones).

Chondroblastoma is a tumor that is rarely located in the pelvis; its more frequent location is the triradiate cartilage. X-rays show a well defined lytic lesion with or without sclerotic margins; it may be 1-6 cm in diameter. Mineralization is frequent, but it is often subtle and is better detected with CAT scan. Tumor expansion could increase the chance of aneurysmatic bone cyst formation. In the MRI T2-weighted images there is variability in signal intensity, which may be due to the presence of hemosiderin, calcifications, and chondroblastic hypercellularity, as well as fluid-fluid levels in the cases associated with aneurysmatic bone cyst.<sup>5</sup> Similar images are seen in clear cell chondrosarcoma, but chondroblastoma occurs at earlier ages, is smaller than clear cell chondrosarcoma, and is confined to the epiphysis.

Histologic studies report a tissue with high cellularity, discrete granulation and matrix calcification that often resembles chicken wire, associated with numerous multinucleated cells present in 20% of cases. The secondary formation of an aneurysmatic bone cyst may be observed.

The definitive diagnosis is made with histopathology. Treatment of chondroblastoma is strictly surgical so as to contain its dissemination to adjacent soft tissues or joint cavities and decrease the recurrence rate. Surgery consists of an incisional biopsy; it may also be combined with excision, curettage and local adjuvant therapy with liquid nitrogen or phenol, selective arterial embolization and even metal implants. Articular surface reconstruction could be necessary in case of extensive subchondral erosion. Less frequently chondroblastoma may occur with metastatic lung disease; in these cases the nodes have to be completely removed. Chemotherapy is not indicated for the treatment of this tumor.<sup>6-8</sup>

### Clinical case

Male, 17 year-old patient, high school student, catholic, without family history of tumors. The current condition began in May 2004 with pain and functional limitation of the right hip after sustaining direct trauma in the right buttock during a recreational sports training. He was seen by the orthopedic surgeon, who ordered X-rays and observed a tumor lesion in the right ischium as an incidental finding. Puncture

and incisional biopsies were taken in November of the same year and in February 2005, respectively; they both reported an aneurysmatic bone cyst (*Figure 1*). Marginal resection of the tumor lesion was performed in May of the same year; the biopsy also reported an aneurysmatic bone cyst. However, tumor volume increased in the following seven months (CAT) (*Figure 2*) and symptoms increased and led to the use of crutches.

The patient was referred to our center, with the following report: right coxalgia, claudicating gait resulting from the right pelvic limb, decreased ranges of motion of the right hip due to pain in the final degrees, and 15 cm-long wound in the right oblique ischiatic region. The X-rays and the CAT scan showed a large cystic, multiloculated lesion, 156 x 140 x 120 mm in its craneocaudal, cross-sectional and anteroposterior axes, respectively; it caused thinning of the anterior acetabular wall with extension to the pelvic organs in zones II and III of the right hemipelvis, according to Enneking. The physical exam did not reveal any functional alterations of the pelvic organs; it did not show neurologic deficit or malaise, and no inguinal adenomegaly was reported. Laboratory test results were within the normal ranges. Surgery was performed on February 22<sup>nd</sup>, 2006, consisting of the following: In a first stage a right ilioinguinal approach was used, the superficial femoral artery was identified all the way to the bifurcation of the common iliac artery; angiography of the internal iliac artery was performed. The tumor lesion was found to have considerable blood supply; the internal iliac artery was ligated and embolized with small pieces of Gelfoam<sup>®</sup> mixed with water-soluble iodinated solution. A sample of the bone tissue was taken for histopathology. In the second stage a posteromedial approach to the ipsilateral thigh was used, 4 cm away from the inguinal zone; curettage of the lesion was performed and a moderate amount of bloody fluid was drained. Controlled fluid hyperthermia was applied with vapor at 70 °C for ten minutes and then abundant hydrogen peroxide was applied.<sup>9-12</sup> No immediate or late complications occurred during this second surgery

neither locally in the wound nor neurologic or related with rectovesical function (*Figure 3*).

The patient's histopathologic results were as follows: Cell groups with blast appearance, with little cytoplasm and round nuclei, as well as osteoclast-like multinucleated giant cells immersed in myxoid stroma with congestive blood vessels and cystic areas compatible with a chondroblastoma associated with an aneurysmatic bone cyst (*Figure 4*).

The patient's clinical course was appropriate during the first four months, assisted by a rigorous rehabilitation regimen. However, X-rays did not show ossification of the cystic area of the ischium associated with persistent mild pain. On June 20, 2006 bilateral iliac crest bone marrow aspirate was performed with an Aspirex<sup>®</sup> needle. The latter was also applied in the right ischium after draining its blood content, to stimulate bone neoformation.

The patient did not attend follow-up visits for 4 years. He presented on February 16, 2011 asymptomatic, walking independently with a discrete pelvic imbalance associated with insufficiency of the gluteus medius, decreased ranges of motion due to predominance of lateral rotation with 15°, 5° of medial rotation, 20° of abduction, 10° of adduction, 100° of flexion and 5° of extension. Muscle strength and sensitivity of the bilateral pelvic limbs were preserved, with a preserved right cremasteric reflex, and a normotonic external anal sphincter (*Figure 5*).

Radiodiagnostic tests showed a well delimited expansive lesion of the right hemipelvis of heterogeneous density, with osteoblastic predominance covering the femoral head; the bladder was delimited by means of the contrast medium used in the intravenous pyelography (*Figure 6*). The <sup>99m</sup>Tc-MIBI scan did not report any evidence of metabolic alterations, which resulted in the absence of skeletal tumor lesions both locally and distantly (*Figure 7*). The MRI reported a lytic lesion distending the cortices in the ilio- and ischiopubic branches, with a heterogeneous appearance, 112 x 128 x 114 mm in its craneocaudal, cross-sectional and anteroposterior axes, respectively, that could correspond to a chondral lesion with a benign appearance (*Figure 8*).



**Figure 1.** AP pelvic X-ray dated 02/2005. Tumor growth is observed.

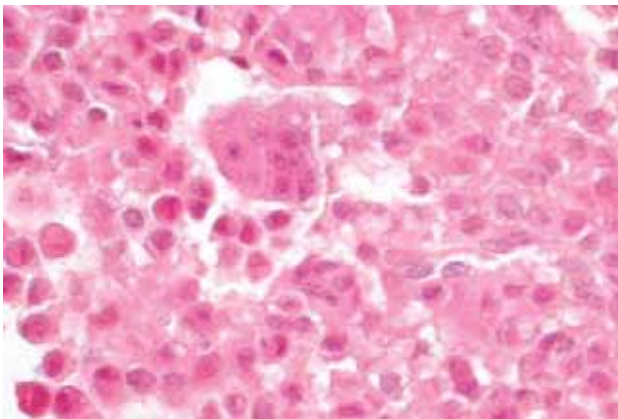


**Figure 2.** Pelvic CAT scan, 3D reconstruction dated 01/11/2006.

The CT-urography reported the following: bladder without tomographic alterations. No data of obstructive uropathy or occupative processes in both kidneys. Vascular structures with normal tract and diameter, only the left hypogastric is observed. A tumor with irregular morphology and undefined borders was observed, with sclerous and intact cortices without images suggestive of a periosteal reaction. The inside of the lesion is heterogeneous, with images with a cystic appearance and mixed densities ranging from 32 to 80 UH which could be blood; with multiple bone neoformation areas and septa without evidence of activity upon passage of the contrast medium. No occupative lesions were seen in the chest CAT scan (Figure 9).



**Figure 3.** Immediate postoperative AP hip X-ray (postembolization of the hypogastric artery).



**Figure 4.** In the cell block it is possible to see the portion of the chondroblastoma composed of giant cells, as well as stromal cells of eosinophilic cytoplasm and round to oval ventral nuclei with nuclear clefts.

## Discussion

Pelvic bone tumors, whether primary or secondary, are difficult to manage, first of all, because they are diagnosed at advanced stages. This is due to: the difficulty in assessing a tumor mass in that area, characterized by a great muscle cover; to the often times intrapelvic growth, and to diagnostic imaging difficulties resulting from unskilled staff. Second,



**Figure 5.** Clinical status of the patient at postoperative year 5.



**Figure 6.** AP pelvic X-ray with contrast medium in the bladder, at postoperative year 5.

because these tumors are difficult to approach and resect. A challenge that all orthopedic surgeons face is trying to avoid injuring the vital neurovascular structures that often times have anatomical variants. Third, marginal tumor resections are not achieved in many cases and they become intralesional with the resulting high recurrence rate of these lesions.

Moreover, pelvic reconstruction surgeries due to damage to the coxofemoral joint (joint replacement, hip transposition, autologous bone grafts, either recycled or homologous, arthrodesis, pseudoarthrosis techniques, use of methyl metacrylate as a filler, pelvic tumor prostheses, etc.) result in numerous complications.<sup>13-15</sup> The use of microwaves and autoclave, among others, is not very effective due to the impossibility of properly exposing the tumor. Last but not least, the urogenital and digestive organs found inside the pelvic cavity may be injured and have sequelae that may result in incontinence and sexual dysfunction.<sup>16</sup>

There are benign tumors with local aggressive, and even metastatic, activity, as is the case of the giant cell tumor and chondroblastoma.<sup>17</sup> This local aggressive activity is expressed not only in the destruction of bone tissue, but in its trend to relapse after proper treatment at a rate ranging from 9 to 30% of the treated cases.<sup>18</sup>

The probable cause of the initial relapse and rapid tumor growth in this patient's pelvis may be explained by what publications on the treatment of chondroblastoma have reported. Relapse is more frequent in the proximal femur region and the pelvis, in epiphyseal locations (less in apophyseal, epiphysio-metaphyseal or metaphyseal ones), in young ages at the time of diagnosis, when time course of symptoms is less than 6 months, and in cases of biologically aggressive tumors.<sup>19-21</sup> However, some authors only mention the history of surgery in the same location as the cause of tumor relapse.<sup>22</sup>

Chondroblastoma has the peculiarity of having secondary pseudotumor formations that «accompany» it throughout its histologic development. They include the aneurysmatic bone cyst, and the unicameral bone cyst, both of

which make surgical management difficult and lead to more complications, as in the case we report herein.<sup>23</sup>

In the pelvis this local aggressive behavior makes treatment even more difficult. The patient reported herein had the aggravating factor of having undergone three prior surgeries and an accelerated tumor growth. Embolization of the right internal iliac artery was performed, considering the history of this procedure in previous papers as treatment for giant cell tumor and pseudotumor lesions one day prior to the marginal resection, as definitive treatment, and also as a palliative option for unresectable malignant pelvic tumors.<sup>24-26</sup> Nevertheless, in this case treatment consisted of adding other adjuvant therapies as controlled hyperthermia with vapor at 70 °C and hydrogen peroxide. The single application of bone marrow in the cavitory areas of the is-

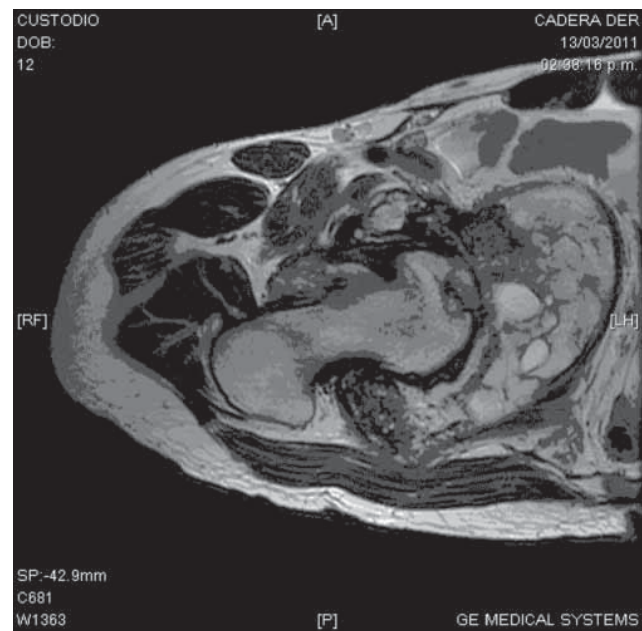
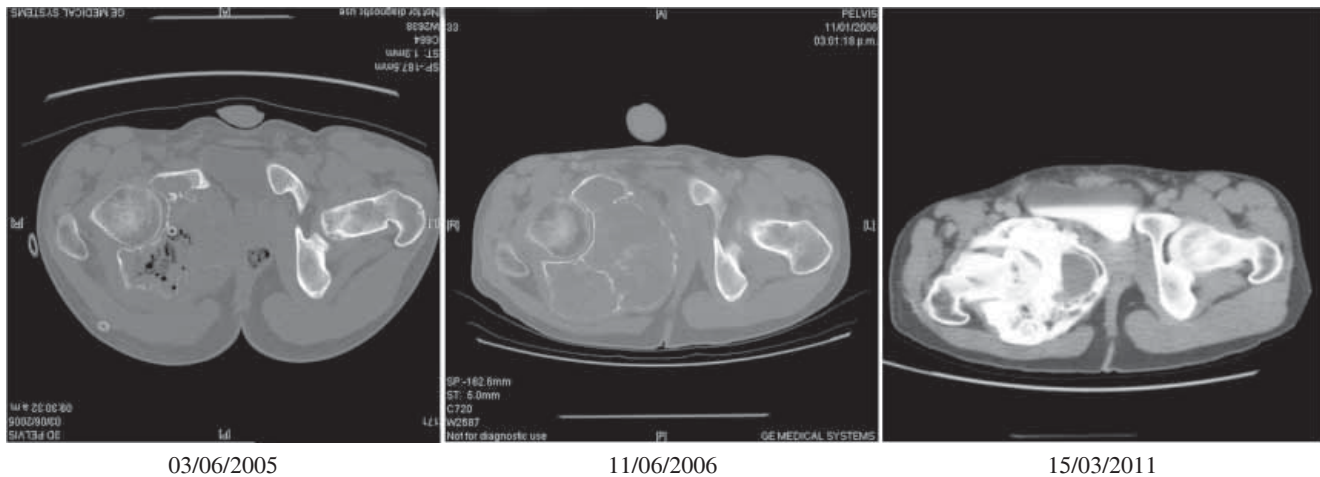


Figure 8. Axial MRI of the right hip, T1 sequence.



Figure 7. <sup>99m</sup>Tc-MIBI scan at postoperative year 5.



**Figure 9.** Comparative axial CAT scan of the same patient showing its time course.

chium contributed to bone tissue neoformation. It should be performed as many times as necessary until appropriate lesion resolution is obtained.

The main purpose of cancer surgery for benign bone tumors is to stop and eliminate the tumor lesion in a certain organ with symptoms or which is at risk. The second purpose, closely linked to the first one, is to relieve pain and maintain function. During the last assessment of the patient reported herein the decrease in the lesion size and the absence of local and metastatic tumor activity were documented from an iconographic perspective (bone scan, CAT, MRI). Besides this, the patient was asymptomatic for bipedestation and ambulation, with a score of 24 according to the functional assessment of the Musculoskeletal Tumor Society (MSTS), which assigns 0 to 5 points to each of the following categories: pain, function, emotional acceptance, use of aids, walking capacity, and posture (maximum score of 30 points).<sup>27</sup>

We think that intralesional resection, associated to local adjuvant treatment (arterial embolization, controlled hyperthermia, tissue peroxidation, autologous bone marrow aspiration and application),<sup>28</sup> was acceptable to treat this patient with chondroblastoma associated with a very large aggressive aneurysmatic bone cyst in the right hemipelvis, with a history of two surgeries and tumor recurrence. The application of autologous bone marrow is based on its osteogenic capacity by means of its osteoprogenitor cells, osteoinduction and osteoconduction. It has been proven that bone marrow transplant to a heterotopic site has the capability of bone induction.<sup>29,30</sup> This patient did not experience any of the complications described in the literature resulting from embolization of the internal iliac artery.

The possibilities of local recurrence and metastasis that may occur with chondroblastoma, even years later, should be considered. We will continue to follow-up the lesion and the lung parenchyma.<sup>31</sup> As reported in the latest pelvic MRI of the patient, moderate coxofemoral osteoarthrotic changes were observed, without osteonecrosis, which overshadow the prognosis of this joint. Symptoms, supported by imaging

studies, will determine the future surgeries that the patient will require, including joint replacement, but with better bone quality in the area and the resulting increased acetabular strength.

When trying to assess a skeletal tumor lesion of the scapular girdle, spine or pelvis, every oncologic orthopedist surgeon should consider that the <sup>99m</sup>Tc-SESTAMIBI scan is the gold standard to rule out malignancy. It is associated with a negative predictive value close to 100%, confirmed with a biopsy.<sup>32,33</sup>

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