

Original article

Unconventional hip arthroplasty for a benign bone fibrous histiocytoma in a pediatric patient after a 15-year course

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ABSTRACT. *Introduction:* Benign fibrous histiocytoma is a rare tumor with very stringent radiologic and histopathologic criteria. It accounts for approximately 1% of all the benign bone tumors. It may be located in both the soft tissues and bone. Its usual location is the shaft, the epiphyseal end of the long bones, the pelvis and the ribs. The age range of presentation is very broad, from 5 to 75 years; it occurs more frequently in young adults. It is rare in children. *Case:* Female, 27 year-old patient who presented at 10 years of age with a tumor in the right proximal femur, which was diagnosed as benign fibrous histiocytoma. She underwent surgical en bloc resection and unconventional arthroplasty with a RIMAG prosthesis. She did fine, had no alterations of implant instability and bone quality. She is developing properly, with symmetry of the extremities and appropriate mobility. *Discussion:* There are only a few articles addressing subtotal and total hip arthroplasty in pediatric ages. There are reports in adolescents ranging from infection to loosening, periprosthetic fracture, revision and, generally speaking, implant failure. However, we did not see this type of complications in this case, and the patient received appropriate management.

Key words: arthroplasty, hip, benign fibrosis histiocytoma, neoplasms, child.

RESUMEN. *Introducción:* El histiocitoma fibroso benigno es un tumor raro con criterios radiológicos e histopatológicos muy estrictos. Constituye aproximadamente el 1% de todos los tumores óseos benignos. Se puede localizar tanto en tejidos blandos como en el hueso. Su localización habitual es la diáfisis, el extremo epifisario de los huesos largos, la pelvis y las costillas. El rango de edad de presentación es muy amplio, abarcando de los 5 a 75 años, presentándose con mayor frecuencia en jóvenes adultos. La presentación en niños es rara. *Caso:* Paciente femenina de 27 años quien inicia a los 10 años de edad con tumoración en región proximal del fémur derecho diagnosticado como histiocitoma fibroso benigno, intervenida quirúrgicamente mediante una resección en bloque y artroplastía no convencional con una prótesis tipo RIMAG. Evolucionando adecuadamente sin alteraciones en la estabilidad del implante, ni en la calidad ósea y su desarrollo es armónico con simetría de extremidades y movilidad satisfactoria. *Discusión:* Pocos son los artículos que hablan acerca de la artroplastía parcial y total de cadera en la edad pediátrica y en adolescentes existen reportes que van desde infección, aflojamiento, fractura periprotésica, revisión y en términos generales de falla del implante, sin embargo en este caso, no se han presentado este tipo de complicaciones y ha resultado ser un adecuado manejo en este paciente.

Palabras clave: artroplastía, cadera, histiocitoma fibroso benigno, neoplasia, niños.

Level of evidence: V (Act Ortop Mex, 2010)

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Introduction

Benign fibrous histiocytoma is a rare tumor with very stringent radiologic and histopathologic criteria, initially described by Dahlin in 1978.¹⁻⁵ It represents around 1% of all the benign bone tumors.⁵

The age range of presentation is very broad, from 5 to 75 years; it occurs more frequently in young adults without gender predilection. This tumor rarely occurs in children; there are very few cases reported in the world literature.^{1,3-6}

It may be located in both the soft tissues and bone. In the soft tissues it may have a superficial or deep location, and very few cases have been reported in bone, as compared with the cases reported in soft tissues.²⁻⁴ Its usual locations are the shaft, the epiphyseal end of the long bones, the pelvis and the ribs. It rarely affects the clavicle, the spine or the skull.³⁻⁵

Its clinical presentation is characterized by being asymptomatic. However, when it occurs with pain, the latter may last from months to several years. A pathologic fracture is not a usual presentation in this type of patients.^{1,3,4}

This lesion has an atypical radiologic pattern. Its radiologic features are frequently similar to those of the non-ossifying fibroma, it is radiolucent with well defined and often times sclerotic borders, without mineralization of the matrix,⁹ which occasionally shows certain degree of expansion. The lesion may have a central or eccentric location in the bone. The bone scan shows moderately increased uptake. The MRI shows an isointense signal with the muscle in T1 potentiated sequences and greater signal intensity in T2.³

Histologically, it is characterized by spindle-shaped fibrous cells that constitute a tissue organized according to a storiform pattern. This tissue contains a variable number of giant cells, hemosiderinic cells and lipid-laden histiocytes; it is thus at times called xanthofibroma or fibrous xanthoma.^{1,2-4}

Concerning the differential diagnosis, from the radiologic standpoint the major entities include non-ossifying fibroma and giant cell tumor, although intraosseous ganglion and osteoblastoma may also be considered. Histologically, it should also be differentiated from a non-ossifying fibroma and a regressing giant cell tumor.^{3,4}

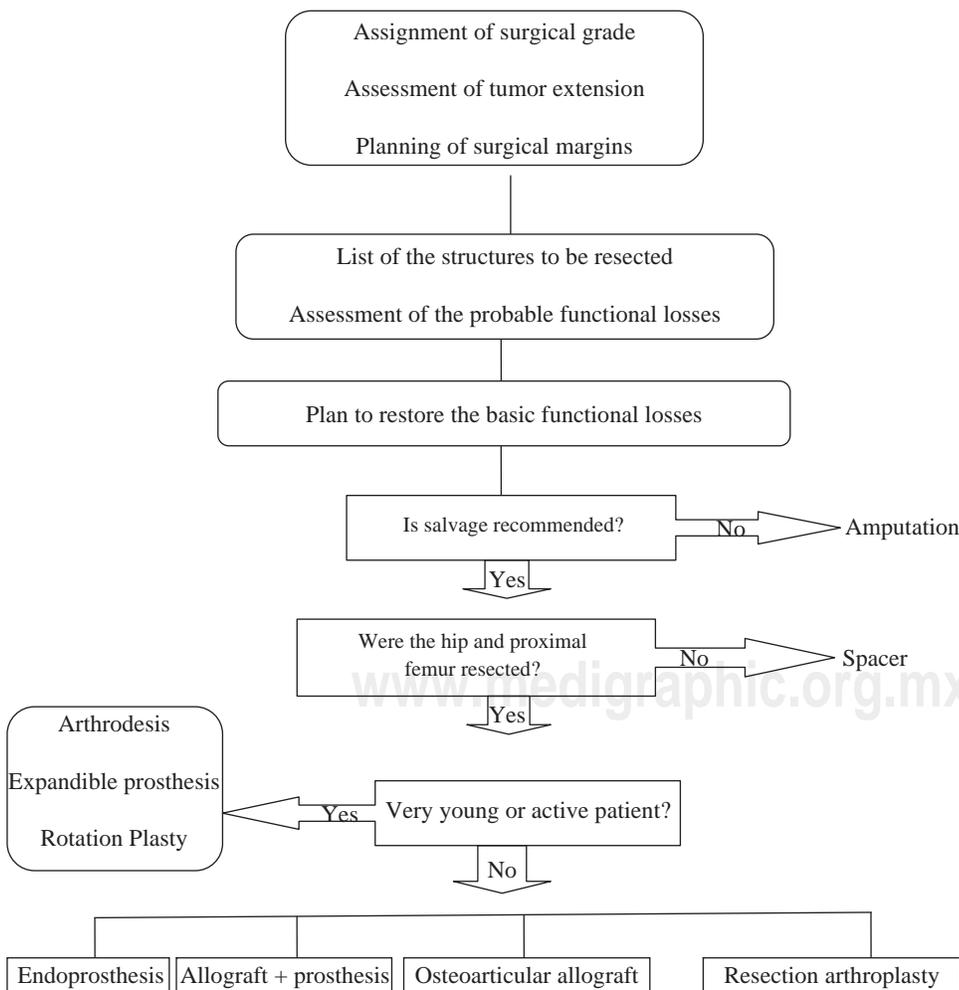


Figure 1. Diagrama de flujo para la reconstrucción del fémur proximal.

Treatment may range from intralesional curettage and grafting to amputation. The lesions that are resectable due to their location should be treated with en-bloc resection.^{2,4} Even though it is a benign tumor, the bone fibrous histiocytoma tends to recur after excision.

Salvage surgery of the proximal femur was first reported in 1943 by Moore and Bohlman, who performed for the first time a proximal femur replacement in a patient with a relapsing giant cell tumor.^{7,8}

The following techniques are available for the reconstruction of the proximal femur:^{7,8}

1. Non-articular intercalary reconstruction
2. Temporary spacers
3. Metal prostheses
4. Intercalary grafts
5. Autologous grafts
6. Articular reconstruction: rotation plasty, expandible prosthesis, femoral endoprosthesis, osteoarticular grafts, endoprosthesis plus graft, resection arthroplasty and arthrodesis.

Johnson and Mankin developed a flow chart for the reconstruction of the proximal femur (*Figure 1*), in which, in the case of young patients, they prefer surgical treatments like arthrodesis, rotation plasties or expandible prostheses.^{7,8}

We describe herein the case of an 11 year-old girl with a diagnosis of a left hip benign fibrous histiocytoma, how

the diagnosis was made, the surgical treatment performed, which consisted of unconventional hip arthroplasty, and her course during the 15-year follow-up period.

The rationale for presenting this case is that this is an infrequent and rare lesion in pediatric ages. Treatment was en-bloc resection of the proximal half of the femur. An unconventional hip prosthesis was used for reconstruction purposes, with a bipolar cup, and a mirror-polished stem locked with bolts. It only required the exchange of the acetabulum for an uncemented component, which allowed for an appropriate musculoskeletal development. The follow-up period was 15 years, something unprecedented in the literature.

Clinical case

An 11 year-old patient was seen at the Bone Tumor Service, National Orthopedics Institute, with a history of a fracture in a previously injured area of the left hip one year back (*Figure 2*).

Her condition began at age 10 and was characterized by a fracture in a previously injured region of the left hip; she was taken to another institution, where she was immobilized for 3 months. No improvement was seen and she was referred to the National Orthopedics Institute, where a biopsy was taken and the diagnosis of benign fibrous histiocytoma was made. She was then scheduled for en-bloc resection of the proximal half of the femur (*Figure 3 a and b, Figure 4 and Figure 5 a and b*) and unconventional hip arthroplasty: an uncemented locked femoral stem and a bipolar cup (*Figure 6*).

The aforementioned surgery was performed in 1995. The patient was followed-up regularly and had unremarkable findings. She started ambulation with weight bearing on postoperative day 4. Four years later she had radiographic evidence of alterations of the acetabular congruence and the decision was made to exchange the bipolar cup for an uncemented cup fixed with 2 screws (*Figure 7*). She has been clinically and radiographically followed-up regularly until today.

She currently does not have any alterations of implant stability or bone quality (*Figures 8a and b*) and her development is harmonic with symmetry of the extremities. She has



Figure 2. Fotografía de cadera derecha.



Figure 3. Fotografía de prótesis no convencional de cadera.

independent gait, without signs of claudication, with 100° hip flexion, neutral extension, 30° of internal and external rotation in supine decubitus, 25° of abduction and 30° of adduction, and a negative trendelenburg (*Figure 9 a and b*). She was referred with a VAS of 0, a WOMAC of 96, a Karnofsky of 90, and an ECOG of 0.

Discussion

There are only a few papers discussing subtotal and total hip arthroplasty in pediatric ages.

There are reports in the world literature of adolescents with hip arthroplasty who sustain from a periprosthetic infection to prosthetic loosening, revision surgery and

overall implant failure; complication rates range from 2% to 29% in patients who underwent salvage of the proximal femur.⁸

The indications for hip arthroplasty in young patients and young adults are very limited. This type of intervention is usually reserved for patients with juvenile rheumatoid arthritis or polyarticular lesions.¹⁰

The most important aspect regarding reconstruction is the soft tissues after the resection of the proximal femur, as they provide stability to the coxofemoral joint. This is achieved by preserving the strength of the abductors by means of myodesis of the gluteus medius to the tensor fasciae latae muscle, a key point for the proper function or failure of the hip arthroplasty.

The benefit of recovering the mobility of the proximal femur joint can have a very important impact on the personal and social development of young patients at this particular stage of their lives.¹⁰

The systems that cause bone ingrowth do not allow performing an appropriate exchange, same as cemented ones, because they alter the bone physiology, particularly in young patients with a good life expectancy.

We think that an important number of cases should undergo this surgery when there is no alternative to preserve the joint. Uncemented systems and locked stems combined with bipolar cups may be used, which would importantly reduce the short-, medium- and long-term complications of other designs.

A stable mechanical fixation of the stem through locking with bolts and mirror-polishing facilitates the exchange, allows preserving the bone bioelasticity and maintaining the bone in better shape for a revision surgery.^{11,12,13}

Reconstruction in this area should be balanced to prevent a lever arm that could lead to the decentering of the stem tip. This is why we try to achieve 40% of reconstruction and 60% of intramedullary stem.¹³

Reconstructing allows for a harmonic musculoskeletal development that avoids the overloading of other joints, which is followed by pain.



Figure 4. Foto clínica de la paciente postoperada.

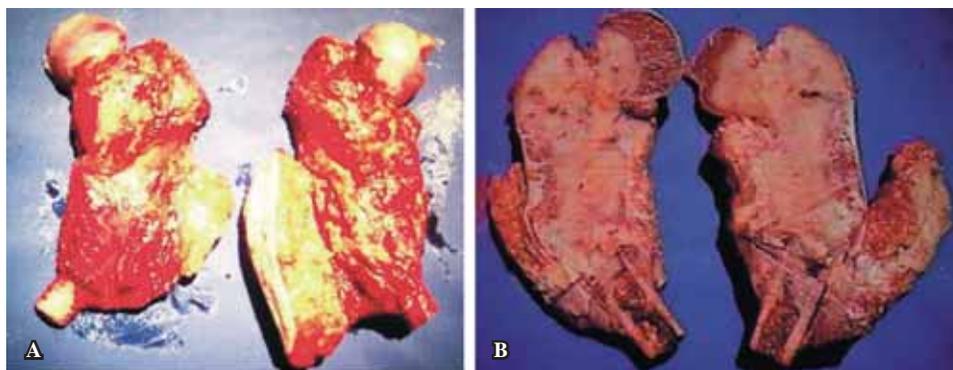


Figure 5. Fotografía de la pieza quirúrgica, **A)** aspecto externo, **B)** aspecto interno.



Figure 6. Fotografía de artroplastía no convencional de cadera: vástago femoral no cementado y copa bipolar.



Figure 7. Fotografía de artroplastía no convencional de cadera con copa acetabular fijo con 2 tornillos.

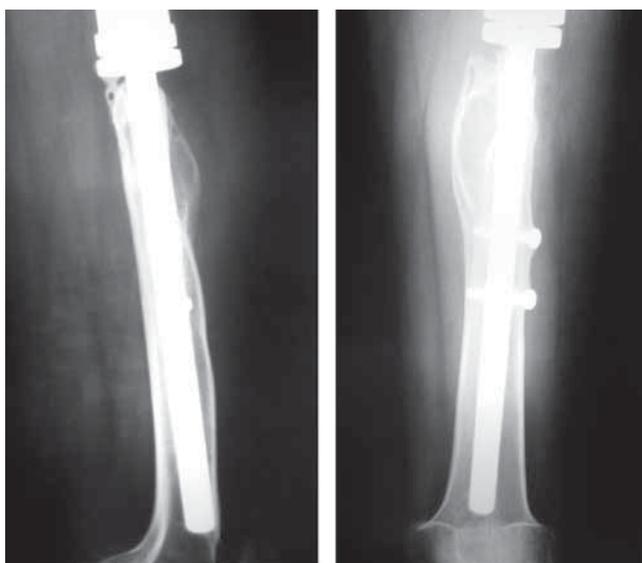


Figure 8. Fotografías de la parte distal del fémur donde no se observan datos de aflojamiento.



Figure 9. Fotografías clínicas en donde se observan los buenos arcos de movilidad.

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