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Case report

Chondrolysis of the hip in an adolescent: clinical and radiological outcomes

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ABSTRACT

Idiopathic chondrolysis of the hip is a rare condition of unknown etiology characterized by progressive destruction of the hyaline cartilage that covers the femoral head and acetabulum. Idiopathic chondrolysis of the hip has an insidious beginning and affects more often female adolescents. Patients report severe hip pain, mobility limitation, and even claudication. This study aimed at reporting one case of that rare disease: an 11-year-old female adolescent with chondrolysis, followed up for three years. Inflammatory activity tests were normal. Imaging tests (radiography, ultrasonography and magnetic resonance) were essential for the diagnosis. The treatment was based on pain control and preservation of the joint mobility, and included low-impact physical activity, non-steroidal anti-inflammatory drugs, and disease-modifying antirheumatic drugs, with good response after 12 months of treatment. Surgery was not necessary.

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Condrolise de quadril em uma adolescente: evolução clínica e radiológica

RESUMO

A condrolise idiopática de quadril é uma condição rara, caracterizada por destruição progressiva da cartilagem articular da cabeça do fêmur e do acetábulo, sem etiologia conhecida. A CIQ tem início insidioso e acomete com maior frequência meninas na adolescência. Os pacientes apresentam dor intensa em quadril, restrição de movimentação e até claudicação. O objetivo do trabalho foi demonstrar um caso dessa doença rara: uma adolescente de 11 anos de idade, com condrolise, em acompanhamento por três anos. As provas de atividade inflamatória eram normais. Os exames de imagem (radiografia, ultrassonografia e ressonância magnética) foram essenciais para o diagnóstico. O tratamento baseou-se no controle da dor e preservação da mobilidade articular, incluindo atividades físicas de baixo impacto, anti-inflamatórios não hormonais e droga modificadora de doença, com boa resposta após um ano de tratamento. Intervenção cirúrgica não foi necessária.

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Palavras-chave:

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Introduction

Idiopathic chondrolysis of the hip (ICH) is a rare condition of unknown etiology characterized by narrowing of the joint space caused by the progressive destruction of the hyaline cartilage that covers the femoral head and acetabulum.¹

In the literature, the description of ICH in the pediatric age group is rare, being mostly restricted to case reports.²⁻⁶ Chondrolysis can occur as an apparently idiopathic event or be secondary to other hip pathologies.⁷

The most common causes of secondary chondrolysis are as follows: prolonged immobilization; neoplasias; Legg-Calvé-Perthes disease (avascular necrosis of the femoral head); trauma; septic arthritis; juvenile idiopathic arthritis (JIA); Stickler syndrome; and slipped upper femoral epiphysis.

Idiopathic chondrolysis of the hip most frequently affects female adolescents (80%) of Asian or African ethnicity, and is more commonly monoarticular (60% affects the right side).^{3,8} Bilateral ICH occurs in 5% of the cases.^{3,8} It is clinically characterized by severe pain in the hip, knee or the entire lower limb, mobility limitation, and claudication. Shortening of the limb might result.

The differential diagnosis of ICH is difficult to be made, mainly with JIA. However, ICH lacks systemic symptoms and has normal laboratory findings (hematological, microbiological, immunological, and acute phase markers).⁷ In addition, the sole involvement of the hip joint is not frequent in JIA.

In juvenile ICH, the radiological images are useful to exclude secondary causes.³ Magnetic resonance imaging plays an important role in the diagnosis and follow-up of that disease.^{8,9}

Some authors have speculated on a genetic and hormonal (girls) etiology, but such hypotheses have not yet been confirmed.^{10,11} Morrissy et al.¹² have suggested that ICH, similarly to slipped upper femoral epiphysis, could represent a seronegative type of immune response, because its joint space narrowing is similar to that of JIA.¹² Other authors have demonstrated that antibodies and immune complexes in the synovial fluid could play an important role in the development of chondrolysis.¹³⁻¹⁶ Thus, although the treatment is still

debated and literature evidence lacks, non-steroidal anti-inflammatory drugs and disease-modifying antirheumatic drugs are used. The use of biologics, such as anti-TNF-alpha agents, has been limited to a case report.¹⁰

In severe cases, when there is no response to clinical treatment, cutaneous traction can be used, in addition to surgery. The major indications of those procedures are improvement of pain and of the hip range of motion, and correction of the deformity.¹⁰ The surgical treatment includes capsulectomy with or without psoas and/or adductor tenotomy, hip arthrodesis and arthroplasty, but the results are not promising.^{11,17} Physical therapy is an important measure in the treatment. According to the literature, remission occurs in 54% of the cases.⁹

Because of the scarcity of reports on ICH in the pediatric age group, we describe the case of an 11-year-old female adolescent.

Case report

The patient is an 11-year-old white female, complaining of pain in the left hip and claudication after physical exercise for one month and a half. She denied infections and/or trauma. She used a non-steroidal anti-inflammatory drug for one month with no improvement. She also denied involvement of other joints. The osteoarticular exam revealed pain and limitation to external and internal rotation of the left hip joint and limping gait. Her laboratory tests showed normal complete blood count, erythrocyte sedimentation rate of 10 mm in the first hour, normal C-reactive protein, and a negative antinuclear antibody result. The diagnostic hypothesis of chronic arthritis of the left hip was suggested. Naproxen (500 mg/day – 12 mg/kg/day) was introduced and slit lamp examination performed, resulting normal. The tuberculin skin test was negative.

The hip radiography (postero-anterior and Lauenstein views) showed mild joint space narrowing and mild osteopenia to the left (Fig. 1). Ultrasonography of the hip showed synovial thickening and joint effusion to the left. On scintigraphy, increased enhancement of the left hip was observed. Magnetic resonance imaging showed joint effusion in the left hip, a small area of 8 mm of hyposignal in T1 and hypersignal in T2 compatible with subchondral edema related to inflammatory process, with no cartilaginous lesion (Fig. 2).

The diagnostic hypotheses were as follows: JIA; avascular necrosis; and chondrolysis. There was no response to naproxen, which was replaced with indomethacin (50 mg/day – 1.2 mg/kg/day). After one month with no improvement, oral methotrexate (15 mg/week – 0.25 mg/kg/week) was introduced. Indomethacin was maintained, and motor physical therapy and swimming were initiated.

After five months, the patient returned to consultation with persistent pain, and reported not using methotrexate for two months. The physical therapy and swimming were maintained. Her physical exam was unaltered. Three months after reintroducing methotrexate, the patient returned to consultation reporting pain on physical exertion and claudication after that. The methotrexate dose was increased to 20 mg, once a week, subcutaneously (0.4 mg/kg/week), the physical therapy and swimming were maintained, and new tests requested. Six months after reintroducing methotrexate, the patient was



Fig. 1 – Initial frontal radiography of the pelvis (Lauenstein, frog view). Osteopenia of the left hip and mild joint space narrowing can be seen.

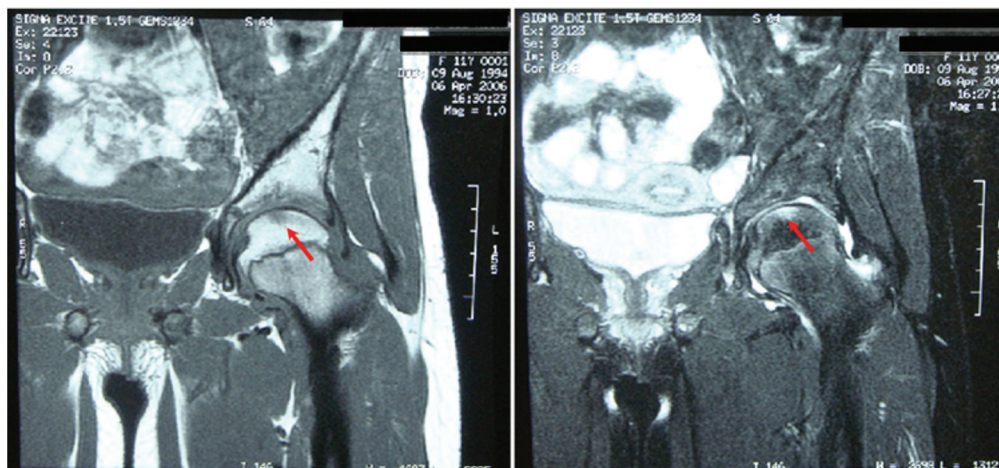


Fig. 2 – Initial magnetic resonance imaging of the left hip. Coronal plane, T1- and T2-weighted fast spin-echo sequence showing moderate joint effusion and a small area with hyposignal in T1 and hypersignal in T2 (arrow), compatible with subchondral edema.

asymptomatic, with no claudication. On physical exam, mild limitation of her left hip mobility was observed.

Twelve months later and 24 months of disease progression, the patient, on regular use of methotrexate, remained asymptomatic with no limitation of her left hip mobility. Her hip ultrasonography was normal. Her left hip magnetic resonance imaging showed mild lateral subchondral thinning, with no edema, and small effusion to the left in T2, showing an improvement as compared with the previous exam. Slow and progressive reduction in the methotrexate dose was initiated. After three months, the patient returned to consultation, with no complaints and denying claudication; her physical exam showed no changes. After four more months with no complaints, methotrexate was suspended. The patient is well. After a 12-month follow-up with no medication, her left hip magnetic resonance imaging showed a small amount of synovial fluid and subchondral and coxofemoral thinning in the posterosuperior portion of her hip (burden area), in addition to mild subchondral edema.

Discussion

We report the case of a female adolescent with chondrolysis, whose initial complaint was chronic pain in the left hip with claudication. She reported no history of trauma, and had limitation of her hip mobility and normal laboratory tests. After receiving methotrexate and undergoing physical therapy, she improved. Her disease is currently under remission with no medication.

According to the literature, ICH usually manifests as hip pain and/or radiated pain to the knees.² The diagnosis is clinical and radiographic.¹ Our patient had monoarticular involvement of the hip, as frequently described in the literature.⁶ In addition, hers is the most frequently reported joint impairment in a female adolescent.³ Functional limitation and limb shortening might lead to claudication, if the diagnosis and treatment are delayed, which was not the case of our patient.^{4,12}

The patient's radiological findings were joint space narrowing and osteopenia. The following radiographic changes of ICH are described in the literature: joint space narrowing; acetabular protrusion; subchondral cysts; joint erosion; premature closure of the growth plate; and lateral increase of the femoral head.^{3,6,8,9,12}

The patient's ultrasonography of the hip showed synovial thickening and joint effusion. The magnetic resonance imaging findings of ICH included early subchondral edema, joint effusion and bone marrow edema; on the progression, focal loss of the cartilage, muscle mass loss and acetabular and femoral remodeling occur.⁷⁻⁹

The diagnosis of ICH is difficult, requiring the exclusion of inflammatory diseases that progress with monoarthritis.^{4,8} The most important differential diagnosis is with JIA, the most frequent chronic arthritis of childhood. However, JIA is hardly ever restricted to the hip, usually affecting other joints.¹⁸ In addition, the patient had neither laboratory test changes (acute phase markers or presence of autoantibodies) nor extra-articular manifestations, such as iridocyclitis. In JIA, the magnetic resonance imaging shows hypervascular synovial thickening (synovial enhancement), reflecting intense inflammatory activity.⁸ Avascular necrosis of the femoral head, epiphysiolysis and neoplasia were excluded based on imaging tests. Trauma as a cause of chondrolysis was also ruled out because of the long duration of the symptoms and progression of the findings. Infectious causes, such as tuberculosis, should also be excluded.

Van der Hoeven et al.⁴ have reported the presence of anti-nuclear antibodies, immune complex deposition and immune disorders in some patients, physiopathological factors of ICH similar to those of JIA. That fact can justify the treatment with drugs usually used to treat JIA, although the literature lacks evidence of good response.

The treatment is based on controlling the progression of the disease and its symptoms, for whose relief non-steroidal anti-inflammatory drugs are recommended. Disease-modifying antirheumatic drugs, such as methotrexate, are used in

the absence of response to anti-inflammatory drugs, as was the case of our patient.¹⁰ Physical therapy and low-impact physical activities are additional measures that should be associated to drug treatment. Despite the poor adhesion to treatment initially, our patient had a good response in the nine months following methotrexate reintroduction. Increasing the methotrexate dose was necessary to achieve that good response. There was complete reversion of the findings, similarly to that reported in other studies.^{2,10} We believe that the early diagnosis and treatment, and the rapid institution of physical therapy were important factors for success.

In addition, the favorable clinical and radiological outcomes prevented the need for surgery. In a case series of 14 adolescents with chondrolysis, about 70% required surgery.¹⁹ Of the 14 adolescents assessed, 4 (28%) had a poor outcome.¹⁹

Idiopathic chondrolysis of the hip should be considered in the differential diagnosis of the monoarticular involvement of the hip. Sequelae, such as an expressive reduction in the joint cartilage or changes in limb size, can be prevented with early treatment.³

Conflicts of interest

The authors declare no conflicts of interest.

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