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Quickly progressive amyotrophy of the thigh: An unusual cause of rapid chondrolysis of the knee

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## **Abstract**

While rapidly destructive OA is more recognized in hip, we report the case of a 50-year-old woman who presented a rapid chondrolysis in the patellofemoral joint in a context of rapid loss of muscular strength. She had arthralgia, myalgia and proximal muscular deficit of the limbs. Creatine phosphokinase level was elevated and electromyogram exam showed a myogenic syndrome. Neither immune nor visceral disease was highlighted. As we suspected a polymyositis, we started corticosteroids and physiotherapy, then methotrexate and intravenous immunoglobulin. Concomitantly to the worsening of the muscular deficit and atrophy of hamstrings, she developed a persistent and disabling knee pain. Initial radiographs and magnetic resonance imaging (MRI) showed only a patellofemoral dysplasia and tiny cartilage damages. Because of aggravation of myalgia, we treated by mycophenolate mofetyl then rituximab. One year later, the knee remained painful and swollen. MRI showed signs of advanced osteoarthritis including an important loss of cartilage with an atrophy of hamstrings. Several articular corticosteroids injections were done. In the same time, the evolution of the muscular disease was unusual. Another histological analysis of muscle has highlighted a genetic myopathy due to mutation of calpain. Immunosuppressive treatments were stopped and a total joint replacement was performed. We show for the first time a case of rapid chondrolysis of patellofemoral joint related to a severe genetic myopathy.

Reduced quadriceps strength or volume is associated with patello-femoral osteoarthritis (OA) (1,2). Rapidly destructive OA is a recognized entity in hip but may occur in knee OA (3). Here, we report a case of a rapid chondrolysis in the patellofemoral joint of a patient with rapid loss of muscular strength due to a severe myopathy.

A 50-year-old woman had myalgia, arthralgia, and proximal muscular deficit of the limbs from one year before the first visit. The rate of creatine phospho kinase (CPK) was 2856 UI/L (normal 10-160 UI/L). A myogenic syndrome was found at electromyographic exam. The antinuclear, antinuclear ribonucleoproteins, anti-synthetase (anti-J01, PL-7, PL-12, OJ, EJ), anti-SRP, anti-Mi2, anti-RNP, anti-Ro52, anti-PM-Scl, anti-Ku, and anti-3-hydroxy-3-methyl-glutaryl-CoA (HMGCoA) reductase antibodies were negative. The histology of deltoid displayed necrotized fibers, complement (C5b9) deposits on sarcolemma of some fibers and sparse inflammatory cells. No over-expression of HLA class I was observed and all the fibers were decorated in immunohistochemistry for the tested structural proteins, including dysferlin and sarcoglycans. Investigations did not identify any neoplasms nor any visceral lesions. Since a polymyositis was suspected, we started corticosteroids (1 mg/kg/day then tapered) and physiotherapy. Because of worsening of the muscle deficit and high CPK level (around 1400 UI/L), subcutaneous methotrexate until 20 mg/week and intravenous immunoglobulin were added. Concomitantly, corticosteroids dosage was increased at 1 mg/kg/day.

Two years later, she developed pain in knees with effusions limiting walking. Clinical examination revealed an atrophy of hamstrings, adductors and gluteal deltoids. The analysis of the joint fluid showed  $<100$  leucocytes/mm<sup>3</sup> in each knee. The initial radiographs showed a bilateral patellofemoral dysplasia without any joint space narrowing. Six months later, magnetic resonance imaging (MRI) examination of the right knee showed very tiny patellar cartilage damages corresponding to an early OA (Figure 1). Five corticosteroid injections in right knee and 3 in left knee (cortivazole and triamcinolone hexacetonide) were performed in the first 18 months with transient efficacy.

Fourteen months after methotrexate initiation, because of the aggravation of myalgia, muscular deficit and persistent increase of CPK level at 1615 UI/L, mycophenolate mofetyl (2 g/d) was added but rapidly stopped because of digestive intolerance, and replaced by rituximab (1000-mg infusion on days 1 and 15) without efficacy.

Nine months after the first knee symptoms, pain dramatically increased and the walk was possible only with a cane. One year after the first imaging evaluation, radiographs showed an almost complete joint space narrowing of patellofemoral joint. MRI showed ulcerations of patellar cartilage and a major atrophy of hamstrings with a normal quadriceps (Figure 2). Considering the functional impairment, one corticosteroid injection (triamcinolone hexacetonide) and one isotopic yttrium injection in each knee and a joint lavage of right knee were performed without efficacy.

Moreover, considering the clinical and biological unusual evolution with immunosuppressive drugs, new analysis on a second muscular biopsy was performed still showing isolated necrotized fibers, atrophic fibers and internalized nuclei without inflammation. Muscle western blot showed a decrease in intensity of the calpain band, exhibiting a genetic myopathy due to mutation of calpain. Then immunosuppressive drugs and corticosteroids were stopped, while intensive physiotherapy was maintained. As the different treatments for knee OA being ineffective, a total knee joint replacement was performed 2 years after the first symptoms.

## **Discussion**

We report a case of a rapid chondrolysis of the patellofemoral joint related to a severe myopathy. Our hypothesis is that the uncontrolled myopathy associated with long-term corticosteroid use lead to a severe and rapid amyotrophy and fatty replacement of the hamstring muscles destabilizing the femoro-patellar joint.

Whereas rapid destructive hip OA is well described (4,5), rapid destructive tibio-femoral OA is less known (3). To our knowledge, it is the first reported case of rapid destructive patello-femoral OA. In our patient, the rapid loss of cartilage (in 1 year) suggests a rapidly destructive knee OA according to Ayral's definition (*i.e.*, a loss of tibiofemoral cartilage  $\geq 50\%$  in 1 year) (3).

In our patient, the rapid functional impairment may have led to the dramatic decrease of the thickness of the knee cartilage as shown in tetraplegic patients (6,7). Unexpectedly, we observed an involvement of the posterior muscular groups of the thigh while quadriceps remained normal, while femoro-patellar OA is actually associated to involvement of quadriceps. A possible explanation is that the balance between agonist-antagonist muscles protects joint against excessive constraints (8, 9) and can worsen OA when it is inverted (9, 10). Our patient had a fatty replacement of hamstrings with quadriceps still tonics that could promote a biomechanical unbalance and consequently rapid femoro-patellar chondrolysis.

In conclusion, severe and rapid thigh amyotrophy should be considered as a cause of rapid chondrolysis of the patellofemoral joint emphasizing the crucial role of muscular strength in patello-femoral OA development.

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**Conflicts of interest:** none

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