Atypical femoral fracture on a deformed bone, treated with a multimodal approach. A case report

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Summary

To report a case of atypical femoral fracture (AFF) induced by long course of oral bisphosphonates (BPs), successfully treated with a multimodal surgical and pharmacological approach. We report about a case presenting to our department with an AFF on a deformed femur (previous surgery for developmental dysplasia of the hip), which we treated with “aggressive” surgical treatment (osteosynthesis + structural bone allograft + autologous bone marrow concentrate) and medical anabolic treatment with teriparatide. Complete radiological fracture healing was observed at 3 months. Difficult fractures should be treated in a combined method, using advanced surgical (usually reserved for non-union or periprosthetic fractures) and pharmacological modalities.

KEY WORDS: atypical femoral fracture; multimodal approach; teriparatide; bisphosphonate; bone graft.

Introduction

Bisphosphonates (BPs) are a class of medications used to prevent osteoporotic fractures. Their mechanism of action involves decreasing bone turnover and increasing bone density, and theoretically improving resistance to fracture.

However, suppressed turnover and the lack of targeted remodeling of the microdamage can also make bones more susceptible to fracture, therefore BPs have been implicated as a contributing factor in low-energy femoral fractures (1). After Odvina’s studies (2, 3), the association between atypical femoral fractures (AFFs) of the subtrochanteric and diaphyseal section and long-term BPs treatment has become accepted. Nonetheless, the causative role of BPs in AFFs pathogenesis remains a matter for discussion (4).

A Task Force of the American Society for Bone and Mineral Research (ASBMR) developed the first case definition of AFFs in 2010, dividing the characteristics into major and minor features and differentiated between complete and incomplete AFFs, while in 2013 they revised the case definition (5). The incidence of AFFs can vary from 0.6% of femoral shaft fractures to 5% (6, 7).

Once the diagnosis of AFF has been made, BPs must be stopped. Internal fixation with intramedullary nailing is the mainstay of treatment for complete AFFs. Prophylactic fixation can also be recommended for impending AFFs, particularly those with extensive cortical defects and pain and/or marrow edema on magnetic resonance imaging, which can lead to delayed healing, non-union or to a complete AFFs (8). We report the case of a 69-year-old woman who presented an AFF which was treated with open reduction and internal fixation (ORIF), structural allogenic bone grafting and autologous bone marrow concentrate (BMC) plus pharmacological anabolic therapy.

Case report

In 2016, a 69-year-old woman with a body-mass index of 24.3 presented to Our Emergency Department with a non-traumatic fracture of the left proximal femur (Figure 1). She reported her left leg slipped away while she was sitting down. She have been suffering of poorly localized thigh pain since the previous month. She had been treating with oral BPs (ibandronate) and cholecalciferol for 10 years, no history of glucocorticoids and proton pump inhibitors assumption was reported. Her past medical history included scoliosis and corrective surgery for developmental dysplasia of the hip (DDH) at the age of 20. In 2015, few months before the AFF, a dual-energy X-ray absorptiometry (DXA) was performed, showing a lumbar (L1-L2) T-score of -1.9 while it was impossible to check the femoral necks because of the DDH (bilateral iliac dislocation with superimposition of the pelvic bone to the femoral neck). All the major features of an AFF were present: subtrochanteric fracture, no history of trauma, transverse orientation of the fracture, no comminution, with localized periosteal and endosteal thickening of the lateral cortex. Further minor features were also present, including cortical hypertrophy (with almost complete obliteration of the medullary canal) and prodromal pain; also, prolonged BPs therapy was present at history...
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Complete blood tests were run, including hepatic and renal function, bone turnover markers, thyroid function and acute phase reactants to rule out any other underlying condition. From the characteristics of the fracture and the negative results of blood tests we diagnosed an AFF occurred in a patient treated with long course of oral BPs. According to guidelines, the treatment with BPs was interrupted (5). From a surgical point of view, fracture fixation with an intramedullary nail (that is the treatment of choice of such a fracture) was to our opinion contraindicated because of obliteration of the femoral medullary canal and major deformity due to DDH and previous surgical treatment. Therefore, we opted for ORIF with plate and screws, allogenic structural bone grafting and autologous BMC. We intra-operative obtained bone marrow aspirate from patient’s right iliac wing and peripheral blood sample, we concentrated both to obtain BMC (which is rich in mesenchimal stem cells, MSCs) and a fibrin clot to be used as a biological scaffold (9). After exposure of the fracture through a standard lateral approach to the femoral shaft, we reamed the femoral canal on the two sides of the fracture to improve endosteal fracture healing potential (10). We prepared a 16-cm long structural emidiaphyseal graft from a fresh-frozen allogenic femoral diaphysis and we located it on the medial aspect of the femur to reinforce the medial wall; the fracture was than fixed with plate and screws on the lateral side. Afterwards, we applied the fibrin clot with BMC in the fracture’s site and in between the structural bone graft and the host bone. Our program of treatment reckoned on medical treatment as well, under prescription of our Center for Osteoporosis. Post-operative course was uneventful. Patient was dismissed to home with non-weight-bearing for 4 weeks in a hinged knee-brace, while mobilization and muscular strengthening exercises of the hip and the knee were allowed. Subjective, clinical and radiographic evaluation were performed at 1, 2, 3, 6 and 12 months after surgery. The 36-item Short Form Health Survey (SF-36) was used to assess subjective status, physical examination looking for range-of-motion, muscle strength, point of tenderness, while X-rays of the femur were obtained in anteroposterior and axial views.

At 1-month follow-up (FU) patient had no pain in her left thigh. Physical examination showed complete healing of the surgical wound. Bone callus formation was observed on X-rays at the medial side of the fracture site. Partial weight-bearing was thus allowed with 2 crutches and while maintaining the brace. At the same time, medical anabolic therapy was started with teriparatide 20 mcg per day via subcutaneous injection. At 2-months FU, patient continued to deny any pain and, as evidenced by SF-36 score, she had resumed almost complete performance of the activities of daily living while still reporting difficulties and insecurity in climbing stairs. X-rays showed an increase in volume of the bone callus. Progressive weight-bearing was encouraged, while maintaining the brace. At 3-months FU, X-rays showed complete healing of the fracture and initial osseointegration of the structural allogenic bone graft (Figure 2), the brace was dismissed and total weight-bearing allowed, and antitromboembolic prophylaxys suspended.

At 6- and 12-months FU, X-rays showed a continuous evolution of the healing process, with the fracture line no longer visible on the radiographs at 6-months and a SF-36 score which resumed the same values as before the fracture occurred. At 12-months FU, patient performed also a DXA test with a lumbar T-score of -0.7 (versus -1.9 at previous testing). Patient decided by herself to suspend the assumption of teriparatide at 18 months, however leaving the opportunity of a new prescription of the medicament for further 6 months, if necessary (Figure 3).

Discussion

AFF have emerged as potential complication of long course
require opening of the fracture leaving intact the fracture hematoma and the osteogenic cambium layer of the periosteum (12). In our case we had no other option than to perform ORIF because of the severe anatomic impairment due to DDH and the strongly corticalized diaphyseal canal. Being a difficult case, we opted for a surgical solution (structural allograft + autologous BMC) we usually reserved for the treatment of complex fractures.
for periprosthetic fractures and nonunions (6, 13), as such a procedure extends the operative time, increasing the risk of infection and costs. Moreover, there is now some evidence in literature that medical anabolic treatment with teriparatide can improve fracture healing (14-16), even if it is still controversial (17). With this multimodal approach, we were able to observe complete radiological fracture healing at 3 months from surgery, while usually it takes 6 months for intramedullary nailing and even more for ORIF (1, 4). It is of course difficult to evaluate which of the “single” treatment performed, if surgical (mechanical or biological) or medical, was predominant in getting a rapid healing. It’s well known that a correct surgical approach is the base of a successful treatment, and sometimes it is also important to promote fracture healing using a medical anabolic support. This concept becomes of paramount importance in fractures with a high risk of delayed union or non-union, such as AFFs (18), even if pharmacological therapy can be used only after a correct evaluation of absence of clinical contraindications and always after a correct osteometabolic study of the patient. In our opinion a combined surgical and pharmacological approach is to be considered as a valuable choice of treatment for “difficult” fractures.

References


