Surgical treatment of neglected hip fracture in children with cerebral palsy: case report and review of the literature

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Summary
Case. A clinical case of a 15-year-old cerebral palsy child with a Sandhu type 2 neglected femoral neck fracture is presented. The patient was treated using cannulated screws and cancellous bone graft augmented with mesenchymal stem cells. At 6 months after the surgery complete fracture healing was observed.

Conclusion. To early diagnose this fractures, it is mandatory to perform a comprehensive clinical and radiological evaluation including also a second level imaging. The use of cannulated screws with cancellous bone graft and MSCs is a viable treatment option in these patients.

KEY WORDS: cerebral palsy; children’s hip fracture; neglected femoral neck fractures; mesenchymal stem cells; diamond concept; non-union.

Introduction
Femoral neck fractures in children are uncommon and usually associated with high energy trauma (1, 2), but physically disabled children are at higher fracture risk with an increased incidence of low energy fractures (3) linked to lower bone density (4, 5) and impaired postural control (6). The treatment of neglected femoral neck fractures is still a challenge for the orthopaedic surgeon (7).

Discussion
The main challenges for the orthopedic surgeon treating children affected by CP are spasticity, delay in developmental milestones, contractures of the soft tissue, bone deformities and their consequences on postural and gait control (11). Hip fractures occur more frequently in this population than in non-disabled children (3) and they are associated to a higher rate of complications compared to others injuries, such as AVN, non-union, coxa vara, and premature physeal closure (12). The overall complication rate was higher in Delbet type I and II (13), and in neglected fractures (7). According to
Meyers et al. neglected femoral neck fractures are subacute fractures diagnosed at least 30 days after the injury (14). Sandhu et al. classified these fractures in 3 stages (Table 1) (10). Henderson et al. hypothesized that a misdiagnosis of fracture in CP could be related to the following conditions: 1) non-significant or recognized trauma; 2) communication impairments; 3) minimally displacement or angulation of the fracture (4).

The treatment of neglected femoral neck fractures is challenging and different techniques are available. Valgus intertrochanteric osteotomy, muscle pedicle bone grafting, reduction and fixation with fibular grafting, and reduction and fixation without fibular grafting are among the options reported (15). In our patient presenting a Sandhu type 2, according to Jain et al. (15), the treatment should be an open reduction (allowing fracture refreshing) and internal-fixation with bone grafting or valgisation osteotomy or open reduction and internal fixation with compression screws and free fibular graft. Valgisation osteotomy is one of the most common procedures performed in these patients. The Putti osteotomy showed its reliability in clinical outcome since 1956 (16) that was lately confirmed also by Zinghi et al. (17). However, these Authors suggested the use of Pauwel’s osteotomy with a degree of lateralization in order to compensate the shortening and prevent the Trendelenburg gait (17). The most common valgisation osteotomy performed is generally the Pauwel’s osteotomy with its several modifications. Recently Min et al. reported good mid-term outcomes in patients treated with a Pauwel-Muller’s osteotomy in 11 cases.
of non-union femoral neck fractures. However, the Authors hypothesized AVN of the femoral head as a possible complication (18).

The effectiveness of Pauwel's osteotomy in neglected femoral neck fractures had been largely reported (15, 19-22).

Magu et al. (20) investigating the effectiveness and safety of a modified Pauwel's osteotomy in 10 children observed a bone union in 9 of them recommending their procedure as the treatment of choice for neglected femoral neck fractures in this population.

On the other hand, valgisation osteotomy is technically demanding and presents some limitations.

Kainth et al. supported the use of valgisation osteotomy in neglected femoral fractures only in selected patients, such as those with a femoral neck resorption less than 2.5 cm, absence of posterior comminution and good bone quality. The Authors argued that valgus osteotomy is technically more demanding and might result in intraoperative anterior angulation and difficulties in reduction in case of posterior comminution. Moreover, a low bone quality was associated with a blade plate cutting out. In these cases, the Authors suggested to use cancellous screw fixation and fibular graft (21).

Varghese et al., studying factors affecting valgus osteotomy outcomes in neglected fractures, observed that reduced preoperative femoral neck absorption ratio (<0.5) was associated to non-union, whereas excessive valgus alignment was associated with worse functional outcome (22).

In particular, excessive valgus alignment could be related to persistent limp and early osteoarthritis and might affect the abductor lever length (23). Wu et al., comparing the use of a compression sliding screw with and without valgus osteotomy, observed that the addition of valgus osteotomy was able to correct a limb shortening of at least 1.5 cm, but it was not associated with a higher rate of bone union, and a higher complication rate (24).

Our patient presented a limb shortening of 1 cm, therefore we preferred to not perform the valgisation osteotomy to preserve the abductor lever arm.

Muscle pedicle bone graft (MPBG) is another available option with good outcomes (14, 15, 25-27). Several MPBGs had been described (i.e. quadratus femoris, tensor fasciae latae and gluteus medius) and the choice among them depends on the surgeon preference. Hou et al. showed complete fracture healing in all of 5 cases treated using vascularized iliac bone graft (26). However, MPBG use is technically demanding and requires microsurgical facilities and experience, which are not widely available (27). Although vascularized fibular bone graft might provide a plus for femoral head vitality (28), nonvascularized one seems to be a dependable and technically less demanding choice (23) in cases of neglected fracture Sandhu 1 and 2 (15). In fact, the fibular graft provides mechanical support and a channel for revascularisation (26, 27). Nagi et al. reported a 70% of...
good results and only 4 AVN at 6-year-follow-up in 40 patients treated with a free fibular graft and one cancellous screw (29). However, their use was associated with some complications such as donor site pain, graft fracture, and gait pattern modification (15, 23, 29-31). We preferred to avoid the use of fibular strut graft considering the risk of gait impairment as reported in literature (31).

Several Authors showed good outcomes using cannulated screws associated with both fibular strut graft (29,30,32) and cancellous bone graft (33). Recently, a biomechanical study demonstrated that placing a third cannulated cancellous screw in the calcar provide a stability advantage for femoral neck fractures fixation (34).

Therefore, we planned to perform an open reduction to refresh the fracture surfaces and to fix it using three cannulated screws with a transverse one in the calcar. The recent introduction of stem cells implantation for fracture non-union treatment is part of the “diamond concept” conceived by Giannoudis et al. (35). Marcucio et al. hypothesized that mesenchymal stem cells (MSCs) might influence bone repair through different mechanisms: 1) differentiation into osteoblasts; 2) triggering the division and differentiation of native connective tissue progenitors; 3) modulating cells of the immune system; 4) secretion of trophic molecules that inhibit apoptosis and fibrosis and/or promote angiogenesis; 5) homing to the fracture site through chemokine receptors, such as CCR1, CCR7, CCR8, and CXCR4-6 and other pathways (36). MSCs use is widely accepted for the treatment of AVN of the femoral head (37, 38) and recently their use has been demonstrated to improve intertrochanteric fracture healing (39). The effectiveness of MSCs in neglected femoral neck fractures was tested by Ayoub et al. in a series of 36 patients.

Figure 5 - Intraoperative pictures. In “a” the antero-lateral approach performed in order to refresh and reduce the fracture; in “b” the stem cells concentrate in liquid and gelled physical state; in “c” application of the stem cells.

Figure 6 - Antero-posterior (a) and axial (b) hip radiographs at 6 months after surgery showing fracture healing and no signs of AVN.
between 15 and 50 years (33) that reported an union rate of 94.4% using cancellous screws and MSCs. The RegenExtra-cell® BMC protocol (RegenLab, Le Mont-sur-Lausanne, Switzerland) is a promising technique of MSCs concentration that allows to achieve recovery rates from bone marrow aspirate of 87% of MSCs (40). In our case, we used the MSCs in two different physical states: liquid and gelled by injecting them into the femoral head and filling the fracture gap. Finally, we used cancellous bone graft from the greater trochanter to completely fill the fracture gap. To identify the appropriate surgical treatment of neglected femoral neck fractures in CP children, we suggest that the surgeon should perform a comprehensive clinical and radiological evaluation, including the GMSCF. In our experience, the use of cannulated screws with cancellous bone graft and MSCs is a viable treatment option in these patients.

Informed consent

This case report respects the Declaration of Helsinki, approved by the Ethical Committee of Unit of Orthopaedics and Traumatology, Martiri del Villa Malta Hospital, Sarno, Italy. Parents were asked to carefully read and sign an informed consent.
Table 1 - Sandhu classification for neglected femoral neck fractures (10, 15).

<table>
<thead>
<tr>
<th>Sandhu type</th>
<th>Fracture surfaces</th>
<th>Proximal fragment</th>
<th>Fracture gap</th>
<th>Femoral head</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>regular fracture surfaces</td>
<td>of 2.5 cm or more</td>
<td>less than 1 cm</td>
<td>no signs of AVN</td>
</tr>
<tr>
<td>2</td>
<td>smooth and sclerosed fracture surfaces</td>
<td>of 2.5 cm or more</td>
<td>between 1 and 2.5 cm</td>
<td>the head of the femur still viable</td>
</tr>
<tr>
<td>3</td>
<td>smoothed fracture surfaces</td>
<td>&lt;2.5 cm</td>
<td>more than 2.5 cm</td>
<td>presenting signs of AVN</td>
</tr>
</tbody>
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