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Neglected femoral neck fractures in cerebral palsy: a narrative review

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- Hip fractures are severe conditions with a high morbidity and mortality, especially when the diagnosis is delayed, and if formulated over 30 days after the injury, is termed a 'neglected femoral neck fracture' (NFNF).
- Cerebral palsy (CP) is probably one of the major risk factors for NFNF in Western countries, mainly because of both cognitive and motor impairments. However, considering the high prevalence of fractures in these patients, the incidence of NFNF in this population is probably underestimated, and this condition might result in persistent hip or abdominal pain.
- Several techniques are available for the treatment of NFNF (i.e. muscle pedicle bone graft, fixation with fibular graft, valgisation osteotomy), but most of them could affect motor function.
- Motor function must be preserved for as long as possible, in order to enhance the quality of life of CP patients.
- After discussing published NFNF cases in CP patients and available treatment options, a practical approach is proposed to facilitate the orthopaedic surgeon to both early identify and appropriately manage these challenging fractures.

Keywords: cerebral palsy; hip fracture; motor function; neglected femoral neck fracture; non-union

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Introduction

Hip fracture is a severe condition which is usually due to a fall in old and/or neurologically impaired patients, or due to high-energy trauma, when it is characterized by a striking clinical picture with hip pain exacerbated by passive movements, inability to fully weight bear and high risk of mortality at short–medium term. Surgery is more successful when it is carried out earlier. However, in some patients this fracture is not diagnosed in a timely fashion, particularly in the case of intracapsular hip fracture. When the diagnosis is made at least 30 days after the injury, hip fracture is more appropriately defined as neglected femoral neck fracture (NFNF).[1]

This condition is commonly described in developing countries,[2–7] because of poverty, ignorance and lack of facilities,[8] although it can be observed also in developed countries, especially in polytrauma.[9] Although the frequency of NFNF is unknown, the incidence of this condition is probably expected to decline considering the improvements in healthcare facilities worldwide.[10] As a consequence, the available literature is mostly about case series in non-disabled patients in underdeveloped countries.[2–7] Disabled patients and especially patients with motor and cognitive impairment are at higher risk of sustaining an NFNF in developed countries.[11–15]

Communicative impairment is one of the most relevant factors leading to a delay in fracture assessment in this population,[12,16] because of the difficulties physicians face in obtaining a comprehensive medical history and physical examination.[16] Typically most fractures in cerebral palsy (CP) occur after a low-energy trauma or simple mobilization of the patient,[14,17] increasing the risk of underestimating patients' symptoms.

NFNF are difficult to treat, considering the loss of bone stock that could modify the femoral neck geometry, and femoral head microarchitecture, negatively affecting the union rate.[5] Moreover, high risk of avascular necrosis (AVN) of the femoral head[3,5,10] further influences both treatment options and surgical outcomes.

Therefore, the aim of our narrative review is to discuss published NFNF cases in CP, in order to identify core critical issues in this field and propose an assessment and therapeutic strategy to help the orthopaedic surgeon to appropriately manage affected patients.

CP and hip fractures

Patients with poor mobility and cognitive impairment are at high risk of misdiagnosis of hip fracture.[13] CP is one of the most common causes of NFNF worldwide, considering the high incidence of osteoporosis due to decreased weight-bearing, endocrine alterations, inadequate calcium and vitamin D intake, and the exposure to medications (i.e. anticonvulsants) that adversely affect bone health.[11,18,19] In non-ambulatory CP children, the low bone mass density (BMD) Z-score (–2 *SD*) and post-operative immobilization are risk factors for long bone fractures.[20] Moreover, CP is often associated with a high risk of falling due to postural control and gait impairments that further increase fracture risk.[14] Maruyama et al[14] observed a lifetime prevalence of fracture of 10% for disabled children. Furthermore, cognitive impairment affects 23–44% of CP patients.[21] Considering also the high prevalence of communicative impairment (up to 80%),[21] a comprehensive medical history is difficult to obtain in this population, leading to a high risk of NFNF.

According to Henderson et al[12] non-significant or unrecognized trauma, communication impairment and minimal displacement are the factors that could lead to a misdiagnosis of a hip fracture in CP. Bajelidze et al[16] observed a positive bone scan in more than half of non-communicative CP patients with persistent pain. Furthermore, as showed by Mariani et al[22] and Kenny and Parkinson[23] hip fractures could mimic abdominal pain in the same population. Therefore, it is mandatory to carefully evaluate persistent hip or abdominal pain, both clinically and radiographically[16,24] in these patients.

As an effect of the extreme variability of the clinical picture of CP, and the lack of available evidence, NFNF are even more difficult to treat in this population. Kenny and Parkinson[23] published a case report of a 16-year-old boy with CP able to walk despite a spastic diplegia, although mentally disabled. The patient sustained bilateral spontaneous hip fractures, initially misdiagnosed because mimicking an abdominal pain. The boy was referred to the authors three months after symptoms began. At that time, he was not able to walk and X-rays showed bilateral subcapital hip fractures. Despite the fact that the patient received no treatment, five years after the diagnosis he was unable to walk, but was pain free.

Mariani et al[22] reported a case of bilateral spontaneous hip fractures mimicking abdominal pain. The patient was 24 years old at the time of the fracture. He was a non-communicative CP patient with a Gross Motor Function Classification System (GMFCS) grade II. He was referred to the authors two months after symptoms began and X-rays showed bilateral Garden IV hip fractures. The patient was treated with a simultaneous bilateral total hip arthroplasty (THA) subsequently complicated by surgical wound infection. After debridement and antibiotics therapy he was discharged, and two years after the surgery he returned to pre-fracture functional status.

Toro at al reported a case of an NFNF which occurred in a 15-year-old boy with CP.[24] The patient's motor function was GMFCS grade II and, after a fall from a standing height, he started to complain of groin pain. After a clinical and radiographic examination, the patient was discharged with the diagnosis of hip bruise. However, because of the persistence of groin pain and the worsening of function, such that he could no longer sustain full weight bearing and walking without a walker few weeks later, he was referred to our institution three months after the injury. The X-rays showed a Sandhu type 2 NFNF on the affected side. Authors performed an open reduction and fixation with cannulated screws and augmentation with stem cells and cancellous bone graft. After an individual rehabilitation plan, the patient was pain free and able to walk without aids one year later.

Meyers et al,[1] in their series of 32 cases treated using muscle-pedicle bone grafting (MPBG), were the original authors who defined as 'neglected' a femoral neck fracture diagnosed 30 days after the injury.

The management of a neglected hip fracture in CP patients

NFNF in young patients are considered one of the most challenging femoral neck fractures to treat for orthopaedic surgeons.[10] Sandhu et al[3] classified the NFNF into three different types based on the shape of fracture surfaces, the proximal fragment length, fracture gap width and femoral head vitality (see Table 1 and Fig. 1 for further details). More recently, Jain et al[7] summarized the available evidence in a literature review proposing a guide to treatment choice according to the Sandhu classification and the time elapsed since the fracture. The treatment of NFNF is a mechanobiological problem[25] and in recent decades several surgical techniques have been proposed for its treatment, including valgus intertrochanteric osteotomy (VIO), MPBG, and reduction and fixation with or without fibular grafting (FG).[2–7,25]

Pivotal targets for treatment choice are femoral head vitality and proximal fragment size.[3,7,10] In fact, hip preservation techniques are considered in all cases of viable femoral head,[3,7,10] with a proximal fragment size of almost 2.5 cm.[3] According to Ayoub and Cad, age (younger

Table 1. Sandhu classification

Sandhu type	Fracture surfaces	Proximal fragment	Fracture gap	Femoral head
1	regular	2.5 cm or more	< 1 cm	no signs of AVN
2	smooth and sclerosed	2.5 cm or more	1.0–2.5 cm	head of the femur still viable
3	smoothed	< 2.5 cm	> 2.5 cm	signs of AVN

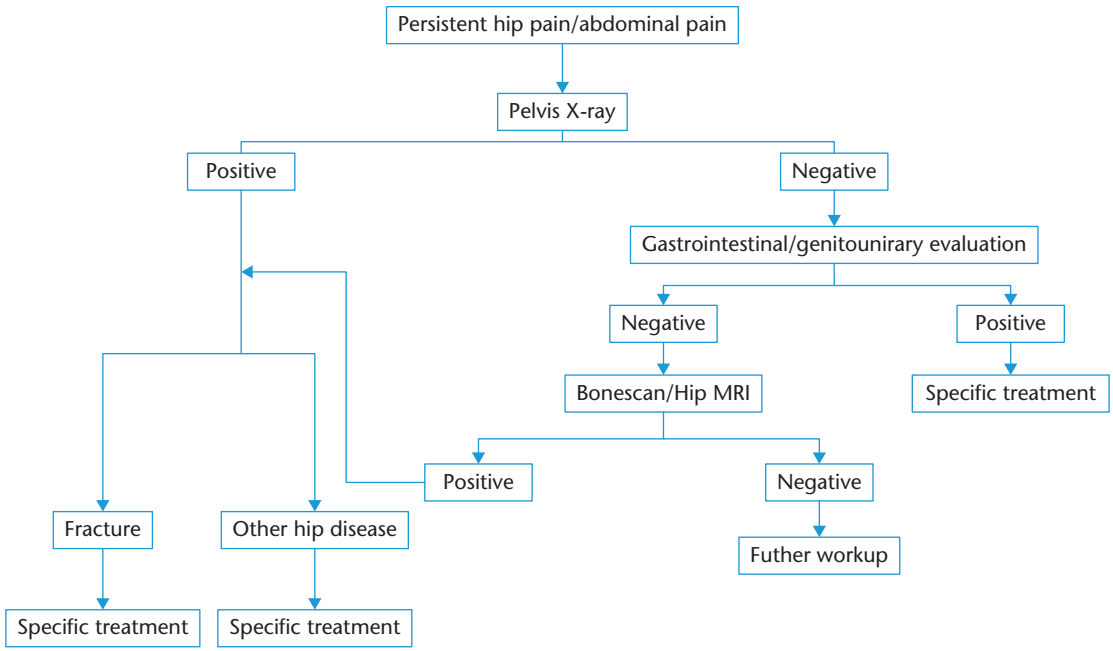


Fig. 1 Diagnostic algorithm for persistent hip/abdominal pain in cerebral palsy.

than 30 years) and post-operative neck-shaft angle (lower than 130°) are the main predicting factors for the best outcomes of surgical joint preservation techniques. On the contrary, time elapsed since the trauma might affect micro and macroarchitectural deterioration of the proximal femur, influencing surgical decision-making.[7]

In CP patients with NFNF it is mandatory that the surgical treatment strategy has to be based on a comprehensive evaluation, including patient's general condition, lower-limb muscle tone, joint contracture and gait impairment,[17] with the ultimate goal of restoring pre-fracture functional status. In CP patients, motor function impairment is commonly classified according to the five stages of the GMFCS.[26,27] Patients able to walk without aids but limited when walking outdoors should be included in the first two stages, whereas those in stages IV and V require assistive technology for most of their daily activities. Stage III is characterized by an intermediate motor impairment.

Patients classified as GMFCS IV or V show the highest risk of hip dislocation,[28,29] that has to be considered in the planning of hip surgical procedures. In fact, even though associated with femoral/acetabular relocation and derotation osteotomy, a joint-preserving technique might be associated with hip instability recurrence in these cases.[30] In these patients, assessment of lower-limb muscle tone, using the Tardieu scale, is required to better manage surgical planning and related outcomes.[31,32]

The surgeon should consider that some of the techniques proposed for the treatment of NFNF could affect the lower-limb biomechanics resulting in worsening of motor function, especially in patients with GMFCS I to III. For example, an excessive valgus alignment after a VIO might negatively affect the abductor lever arm resulting in a persistent limp,[33] whereas the use of fibular graft is associated with gait impairment and ankle instability.[34] Therefore, the surgeon should carry out an adequate risk-benefit analysis, preferring less risky procedures (i.e. fixation with cancellous screws and bone graft). Recently, stem-cell transplant was proposed to treat fracture nonunions with encouraging results.[5,35] Also, our experience in a clinical case treated with cannulated screws, bone graft and stem cells reported good results at one year follow-up.[24]

Total hip arthroplasty (THA) might be the preferred treatment choice in CP patients with persistent hip pain with good long-term survivorship and a complication

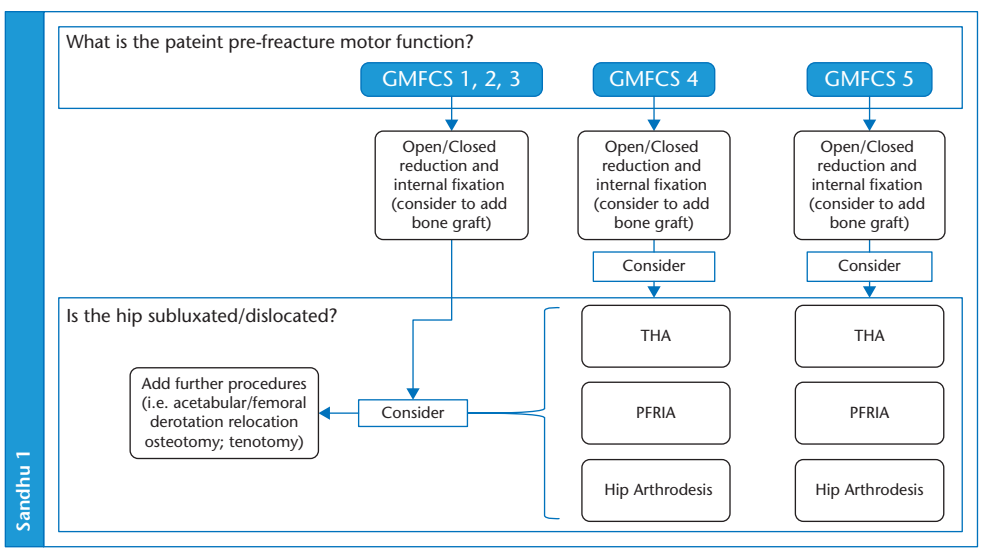
rate comparable with THA in non-disabled patients.[36,37] According to Houdek et al[37] GMFCS stage does not influence THA outcomes. In any case, the surgeon has to consider that performing THA in patients with a neurological disorder has some difficulties to face in terms of pre-operative counselling, altered acetabular and femoral anatomy, dealing with effects of previous surgery to the hip, the need for altered post-operative rehabilitation and a high rate of complications.[38,39] Some technical precautions may help to improve THA outcomes in these patients.[39]

Indeed, the use of dual-mobility cups further reduces the THA dislocation risk in this population[40] and some authors have associated femoral derotational osteotomy with THA in cases of dislocated painful hips.[41] Therefore, hip replacement has been also proposed in ambulatory patients in the more severe GMFCS stages. Mariani et al[22] reported a case of NFNF in CP treated with bilateral THA with good outcomes at two-year follow-up. In Sandhu type 3 NFNF, THA should be a treatment option also in CP ambulatory patients. A salvage procedure such as hip arthrodesis might be an alternative choice, especially in CP patients with normal contralateral hip and without structural and/or functional spine impairments,[42] even if this procedure is associated with high complication rate.[43]

For non-ambulatory patients or for those with a severe impairment of gross motor function (GMFCS V) the surgeon might also consider proximal femur resection techniques. Generally these procedures are indicated as salvage surgery to treat dislocated painful hip and to allow the patients to sit comfortably and to make perineal care possible.[44-47] Proximal femur resection techniques are generally associated with severe complications, whereas the Castle subtrochanteric resection-interposition arthroplasty (PFRIA) seems associated with more reliable surgical outcomes.[48,49] In order to reduce the risk of bone-to-bone impingement when proximal migration of the femur occurs, Silverio et al[50] proposed the interposition of a shoulder prosthesis instead of soft tissues. Kenny and Parkinson,[23] in a case report, scheduled a CP patient with NFNF for proximal femur resection, but the surgery was not performed because of spontaneous pain resolution. According to both literature findings and our expertise, we propose viable options to treat NFNF in CP (Fig. 2).

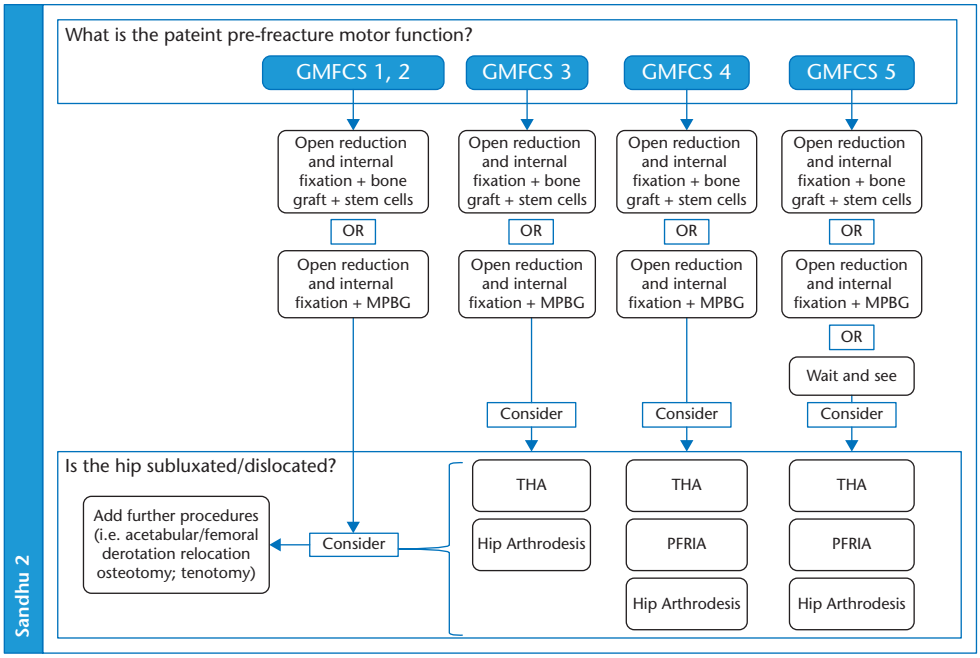
Conclusions

Although hip fracture has a high incidence in patients with CP, diagnosis is frequently missed. In our opinion, hip fracture should be suspected in a case of persistent and/or unexplained hip or abdominal pain in this population to allow early fracture identification. When diagnosis is delayed for over 30 days, this condition is defined as NFNF, and for its treatment in CP patients the surgeon should consider the GMFCS stage, in order to formulate an appropriate surgical plan to achieve favourable outcomes. Surgical techniques that do not affect motor performance should be preferable to preserve ambulatory function. Total hip arthroplasty is a viable option in ambulatory patients with a Sandhu type 3 NFNF. In non-ambulatory patients, resection of the proximal femur might be indicated.



(a)

(continued)



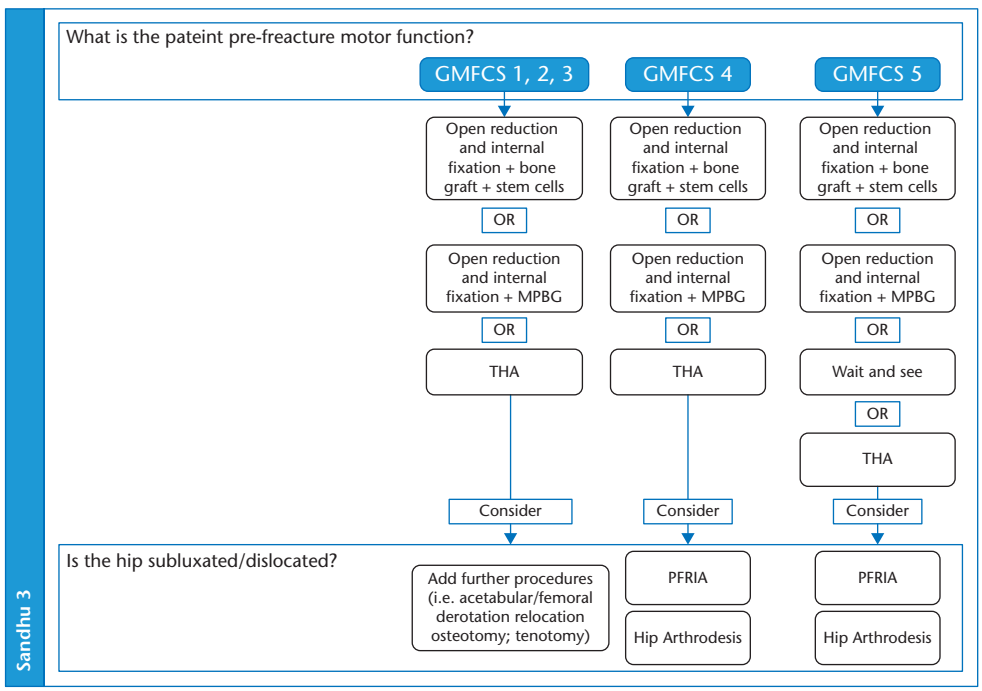


Fig. 2 Comprehensive algorithm for the treatment of neglected femoral neck fracture (NFNF) in cerebral palsy (CP) according to Sandhu type and GMFCS. (a) Algorithm for the treatment of Sandhu type 1 NFNF. (b) Algorithm for the treatment of Sandhu type 2 NFNF. (c) Algorithm for the treatment of Sandhu type 3 NFNF.

Note. GMFCS, Gross Motor Function Classification System; THA, total hip arthroplasty; PFRIA, Castle subtrochanteric resection-interposition arthroplasty; MPBG, muscle-pedicle bone grafting.

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