International Journal of Academic Health and Medical Research (IJAHMR)
ISSN: 2643-9824
Vol. 8 Issue 6 June - 2024, Pages: 84-89

Neurogenic paraosteoarthropathies

Zineb Oudrhiri ;Hajar Dardar; Mohammed Tazi Charki; Hicham Abdellaoui; KarimaAtarraf ;Moulay Abderrahmane Afifi

Pediatric Orthopedic Surgery Department, Hassan II University Hospital, FEZ

Abstract*: Neurogenic paraosteoarthropathies (POAN) are abnormal processes of ectopic neo-osteogenesis that develop near large joints, occurring after central or peripheral neurological lesions as well as in the course of certain comas. They are responsible for joint stiffening and a vicious attitude, which can lead to nerve or vascular complications through compression. Here, we report a case.*

Keywords: Neurogenic heterotopic ossification, multiple trauma, neurological disorders, Management ,surgery

1. Introduction

Neurogenic paraosteoarthropathies (POAN) are characterized by ectopic ossifications that develop near large joints. [1]This process corresponds to ectopic neo-osteogenesis occurring following central or peripheral neurological lesions, as well as in certain cases of coma. [2] The challenge is therefore to quickly detect the disease to avoid various complications or the need for major surgery.

2. Clinical observation

We present the case of a 9-year-old right-handed patient with no notable medical history, who suffered multiple traumas following a road traffic accident. The accident caused a traumatic brain injury with a parieto-occipital subdural hematoma extending to the cerebellar tent, non-surgical, limb trauma with a left femoral fracture,(figure 1) and a lower left eyelid wound. The patient was intubated, ventilated, and sedated upon admission due to neurological criteria. After stabilization of the hemodynamic status, the patient underwent a stable centro-medullary nailing (figure 2) and remained hospitalized in intensive care for 10 months. During hospitalization, the electroencephalogram showed a pattern of diffuse cerebral distress predominantly on the right side. A control MRI revealed diffuse ischemic areas with laminar necrosis in the temporo-occipital region. The patient was extubated and tracheotomized after 1 month of hospitalization. During hospitalization, the patient developed an infection with the presence of Acinetobacter in the protected distal sampling. After extubation, the patient had a Glasgow score of 10. Radiological control of his fracture at 1 month showed no anomalies with the beginning of consolidation. Five months later, the presence of a bone bridge was observed at the level of the right knee (figure3 and 4) and at the level of the hip of the operated limb (figure5and6). The diagnosis of neurogenic paraosteoarthropathy (POAN) was made, and the patient did not present any clinical signs of compression or other complications, hence the decision to perform a bone scintigraphy to evaluate the activity of the osteoma. The decision was not to operate on the patient due to the high risk of recurrence and to wait for the maturity of the osteoma.

3. Discussion

Neurogenic paraosteoarthropathies (NPOA) remain a common concern in medicine, affecting 10% to 53% of spinal cord injured individuals [1-3], and 10% to 28% of TBI cases. [3-4]

NPOA are usually localized around the large joints such as the hip, knee, elbow, and shoulder.

However, a few cases have been reported with a distal development. [5] The hip is affected in 28.7% to 91.6% of cases. [6] In our case, the localization was the knee and the hip.

Pain is the first clinical manifestation in patients who have retained sensitivity [7,8], which is not the case for our patient.

Radiography confirms the diagnosis of paraosteoarthropathy, detects other locations, and tracks their evolution. Computed tomography allows for diagnosis within two weeks of symptom onset, and thus before radiography [9]. Bone scintigraphy with technetium Tc-99m was proposed several years ago as a method for early detection and monitoring of POA maturation before surgery. [10]

However, when scintigraphy is combined with CT scanning (SPECT-CT), the specificity increases and allows for a definitive diagnosis [11]. our patient underwent both procedures.

According to some studies, prolonged coma or persistent vegetative state, as well as an initial Glasgow coma Scale score between 3 and 5 in patients with TBI sequelae, are established risk factors for POAN. [6-12]

A significant correlation between the development of POAN and the occurrence of fractures has been reported in the literature. [2] This risk is significantly increased by surgical intervention such as osteosynthesis. [13] Which is the case with our patient.

In most POAN cases, the main biological parameters are normal. Only the serum level of Alkaline Phosphatase (ALP) can be used for early diagnosis of POAN. Indeed, from the second week after the initial injury, ALP levels begin to rise, reaching a peak of 3 to

4 times the normal level by the 10th week. This level returns to normal around the 18th week. [11] The early phase of care in intensive care units with appropriate rehabilitative management is very important in preventing POAN, especially in patients with known risk factors. [2] Some studies [14] have demonstrated the efficacy of bisphosphonates for their inhibitory action on osteoclastic resorption and bone formation. However, they have been quickly questioned due to the risk of osteomalacia with long-term treatment and the rebound effect upon premature cessation [15] The only curative treatment consists of surgical excision of the POAN when it is established. It can only be considered in cases of significant functional loss or neurological and vascular complications, [1] which is not the case for our patient. POAN can continue to evolve for up to 1 year following the onset of the initial pathology [16]. Their size eventually stabilizes thereafter.

Genêt et al. showed that early surgery can be considered as early as the first year of evolution without increasing the risk of recurrence [17], thus avoiding progression to ankylosis and the occurrence of complications. it was believed that a first excision surgery on insufficiently mature bone would increase the risk of postoperative recurrence [18]. However, it is now known that this assumption is incorrect. Genêt et al. have shown that operating on POAN early, within the first year following the neurological injury, does not increase the recurrence of POAN [17-19]. They further stated that the size of the preoperative POAN, the severity of cognitive and motor deficits in operated patients, and the multi-site nature of the POAN to be resected are not correlated with POAN recurrence [17-19–20].

4. Conclusion :

Neurogenic para-osteo-arthropathy (POAN) results in joint stiffening and deformities that significantly affect function. Early diagnosis through imaging is crucial to prevent complications. Surgical excision is the primary treatment once established.

Figure 1: initial X ray showing a femur fracture

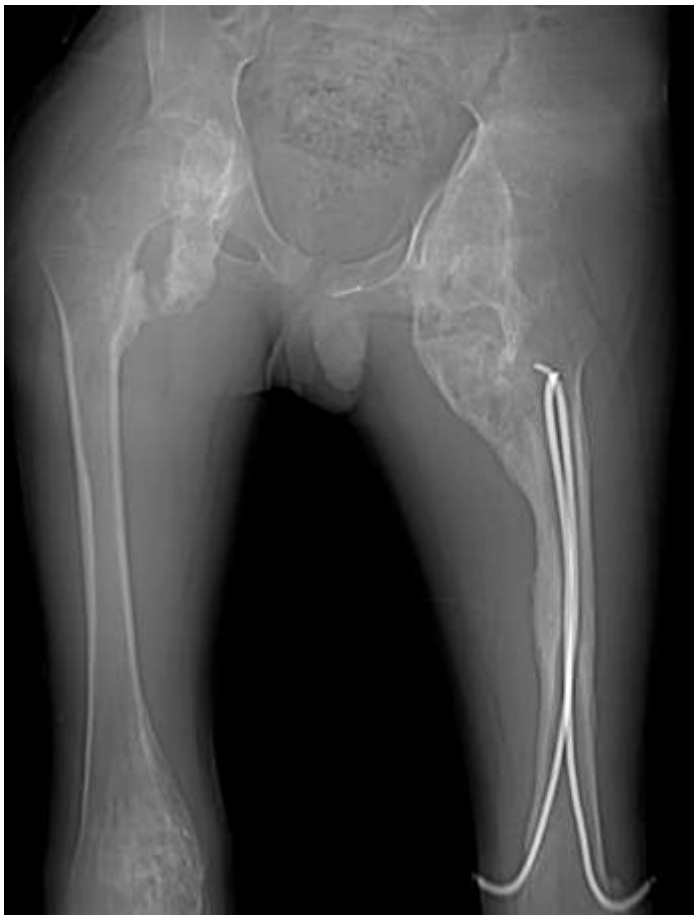


Figure 2: intramedullary nailing and early para-osteoarthropathy

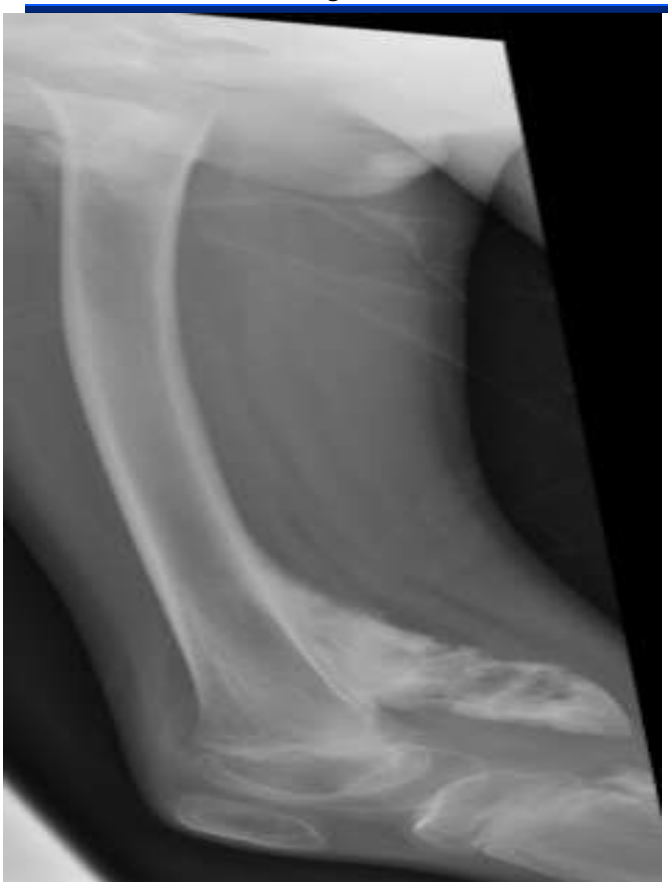
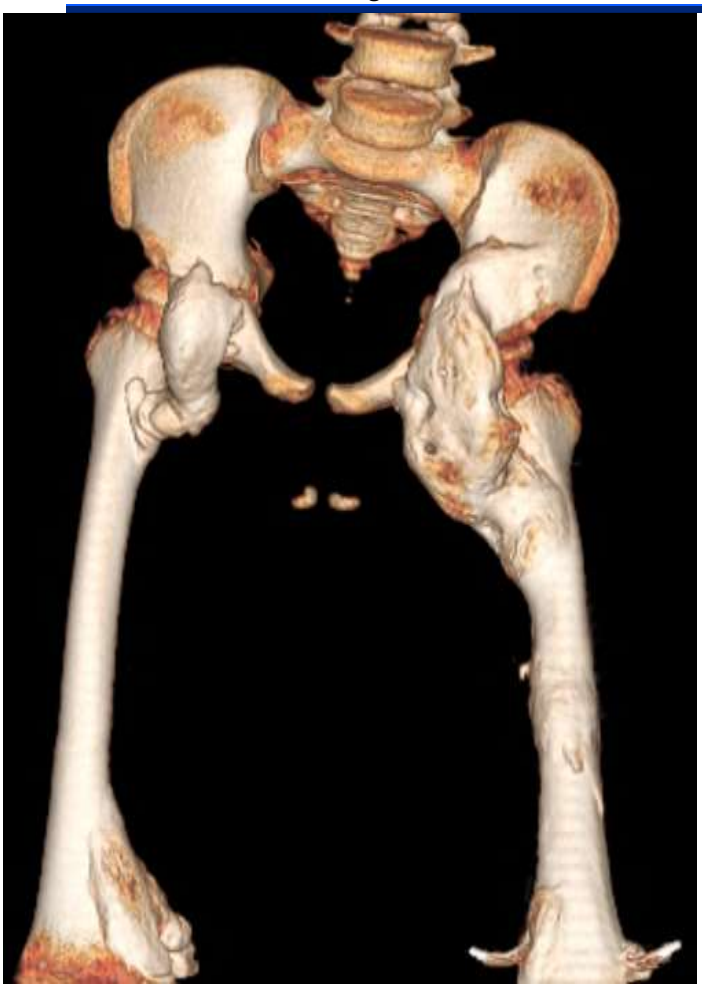


Figure 3: standard X ray showing elbow involvement



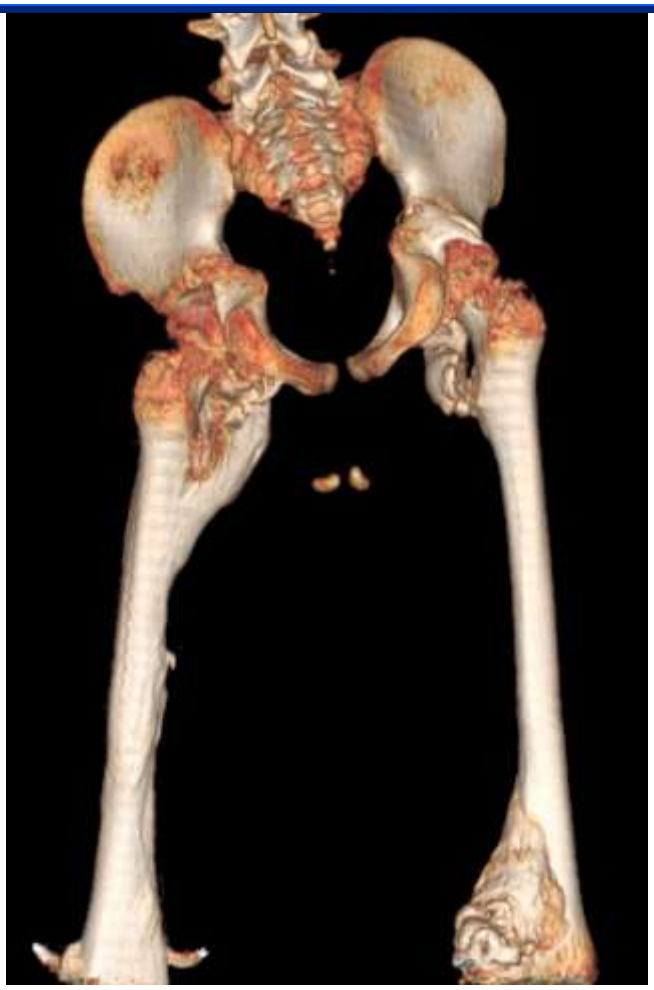


Figure 4 and 5 : anterior and posterior views of a cT reconstruction showing involvement of the hip

References :

1:O.Kharrata,*, M. Ouertatanib, K. Ben Hmidab, M. Mestirib, C. Dziria Neurogenic periarticular heterotopic ossification: A case report Journal de re´adaptation me´dicale 2015;35:42-46
2:Uebelhart D. Les para-oste ´o-arthropathies nerveuses : mise au point a` destine ´e re ´e´ducative. Ann Readapt Med Phys 2002;45:181–7.
3:Isaacson BM, Brown AA, Brunker LB, Higgins TF, Bloebaum RD. Clarifying the structure and bone mineral content of hetero topic ossification. J Surg Res 2011;167:e163–70.
4:Galien P, Nicolas B, Le Bot MP. Heterotopic ossifications re sponsible for vascular compression. Rev Rhum Engl 1994;61: 733–7.
5:Marjorie Salga Inflammation et paraostéoarthropathies neurogènes Thèse de doctorat de l'Université Paris-Saclay Préparée à l'UVSQ École doctorale n°569
6:Flin G, Curalucci H, Duvocelle A, Viton JM. Para-ostéarthropathies neurogènes et traumatisme crânien sévère. Ann Readapt Med Phys 2002;45:517–20.
7:Pelissier J, Petiot S, Benaim C, Ascencio G. Prise en charge des para-oste ´o-arthropathies neuroge `nes (POA) chez le traumatise ´ cranioence ´phalique : e´tude de la litte ´rature. Ann Readapt Med Phys 2002;4:188–97.

8:Chung SY, Oh KK, Kim DJ. Mammographic and sonographic findings of a breast cystic lymphangioma. J Ultrasound Med 2003; 22:307–309.Cited Here |PubMed | CrossRef
9:Ogun GO, Oyetunde O, Akang E. Cavernous lymphangioma of the breast. World J Surg Oncol 2007; 5:69–72 Cited Here
10:akellariou VI, Grigoriou E, Mavrogenis AF, Soucacos PN, Papagelopoulos PJ. Heterotopic ossification following traumatic brain injury and spinal cord injury: insight into the etiology and pathophysiology. J Musculoskelet Neuronal Interact 2012;12:230–40.
11:Ghanem MA, Dannoon S, Elgazzar AH. The added value of SPECT-CT in the detection of heterotopic ossification on bone scintigraphy. Skeletal Radiol 2019. https://doi.org/10.1007/s00256-019-03286-x.
12:Rusdianto Eve, Murray Mary, Davis Jennifer, Caveny Anne. Cases of Atypical Lymphangiomas in Children. Case Reports in Pediatrics. 2014
13:Minocha Prashant K, Roop Lakhan, Persad Rambachan.Cases of Atypical Lymphangiomas in Children. Case Reports in Pediatrics 2014:2014
14:Boufettal Houssine, Noun Mohammed, Hermas Saïd, Naïma Samouh, Marouane Soufia. Lymphangiome mammaire. Elsevier Masson.2010; 20(4): 222-227. Google Scholar
15:Tyler JL, Derbekyan V, Lisbona R. Early diagnosis of myositis ossificans with Tc-99m diphosphonate imaging. Clin Nucl Med 1984;9(5):256–8.
16:Garland DE, Hanscom DA, Keenan MA. Resection of heterotopic ossification in the adult with head trauma. J Bone Joint Surg 1985;67A:1261–9
17:Genêt F, Jourdan C, Schnitzler A, Lautridou C, Guillemot D, Judet T, et al. Troublesome heterotopic ossification after central nervous system damage: a survey of 570 surgeries. PloS One 2011;6:e16632. https://doi.org/10.1371/journal.pone.0016632.
18:Almangour W, Schnitzler A, Salga M, Debaud C, Denormandie P, Genêt F. Recurrence of heterotopic ossification after removal in patients with traumatic brain injury: A systematic review. Ann Phys Rehabil Med 2016;59:263–9. https://doi.org/10.1016/j.rehab.2016.03.009.
19:Genêt F, Chehensse C, Jourdan C, Lautridou C, Denormandie P, Schnitzler A. Impact of the operative delay and the degree of neurologic sequelae on recurrence of excised heterotopic 236 ossification in patients with traumatic brain injury. J Head Trauma Rehabil 2012;27:443–8. https://doi.org/10.1097/HTR.0b013e31822b54ba.
20:Genêt F, Ruet A, Almangour W, Gatin L, Denormandie P, Schnitzler A. Beliefs relating to recurrence of heterotopic ossification following excision in patients with spinal cord injury: a review. Spinal Cord 2015;53:340–4. https://doi.org/10.1038/sc.2015.20