

Case Report

Neurogenic Para-Osteo-Arthropathy in a Drowning Patient with Cerebral Anoxia: (A Case and the Review of the Literature)

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Abstract

Paraosteoarthropathy (POAN) corresponds to the formation of heterotopic extra-articular ossifications, developing in muscle and fibromesenchymal tissue. It is said to be neurogenic when it occurs following an attack on the central or peripheral nervous system. It is frequently described after a head trauma or a spinal cord injury, but much less often after a neurovascular pathology. It occurs relatively early in the resuscitation phase. The formation of these ossifications causes significant pain and this can result in significant stiffening sometimes in vicious positions. Their exact pathophysiology remains unknown despite numerous studies and cases were reported. We present the case of bilateral hip paraosteoarthropathy in a drowning patient with cerebral anoxia.

Keywords: Paraosteoarthropathy; Heterotopic ossifications; Drowned; Cerebral anoxia

Introduction

A POA corresponds to the formation of heterotopic ossifications around a joint. It is always extra-articular, but can come into direct contact with the joint capsule. They are located almost exclusively around the large proximal joints of the limbs: hips, elbows, knees, shoulders. The POANs result in tables of inflammatory pseudo-arthritis which can progress to ankylosis of the joints, the functional consequences of which can be major. Currently, surgical resection of ossifications is the only therapeutic alternative, followed depending on the case of a total arthroplasty, with appropriate rehabilitation given the risk of recurrence and instability of the joint [1]. We report the case of a POAN in a drowning patient with cerebral anoxia with a review of the literature.

Case Presentation

A 28-year-old patient, having spent 3 years in intensive care for three months for acute respiratory distress syndrome following drowning, intubated and put on assisted ventilation for 5 weeks, which presented pains of the two hips more noticed on the right side with stiffness of the right hip and a limitation of the articular movements, At the clinical stage, the examination revealed a vicious attitude of the right hip in flexion of 20°, irreducible, with stiffness in mobilization and inability to walk without help, i.e. a PMA (Postel Merle d'Aubigné) score of 3. The patient underwent a biological assessment which did not reveal any inflammatory syndrome, the rheumatic factors were negative while the serum alkaline phosphatases were at the normal upper limit. Radiography of the pelvis showed a bony bridge between the greater trochanter and the right acetabulum (Figure 1). The CT found a large calcium formation forming a neocalcification between the posterior edge of the greater trochanter and the internal and posterior edge of the acetabulum. The bone scintigraphy showed fairly intense hyperfixations at the level of the newly formed bridges, witness to their evolutionary character.



Figure 1: Radiography of the pelvis showed a bony bridge between the greater trochanter and the right acetabulum.

The patient underwent surgical arthrolysis with resection of the newly formed bridge (Figures 2 and 3) and postoperative physiotherapy. The evolution was very favorable, painless free hip without recurrence after one year (Figure 4).

Discussion

POAs are defined as the formation of lamellar bone in non-bony tissues, particularly muscles and connective tissue, where bone normally does not exist. POA, or heterotopic ossifications, were identified for the first time and described in 1883 by Riedel [2], a German doctor. They were later described as “paraosteoarthropathies”



Figure 2: Intraoperative view of resection of the newly formed bridge.



Figure 3: View of the respected bone bridge.

by doctors Dejerine and Cellier from observations of patients with traumatic paraplegias during the 1st World War. [3] There are 3 recognized contexts of POAs: traumatic, neurological and genetic.

In terms of pathophysiology, most authors raise the hypothesis of the transformation of hematopoietic cells placed in an extra-skeletal situation into osteogenic cells, following periosteal tears secondary to spastic movements in comatose and spinal cord injured patients [4-5].

From a clinical point of view, unless there is systematic screening, the average time between the occurrence of the neurologic lesion and the diagnosis of POAN is 2 months [6], with a variation ranging from 15 days to 1 year [7]. The clinical signs and symptoms of heterotopic ossifications appear between 3 and 12 weeks after the initial lesion. This delay varies according to the etiologies of the POANs. When they are symptomatic, they usually cause a decrease in mobility of the affected joint, and in severe cases complete ankylosis may appear [8]. POANs can also induce local pain and, if localized superficially, symptoms such as edema, erythema and heat. Nerve or venous compression may also occur [7-10]. The sex ratio is similar, but the incidence of POANs is significantly lower in children than adults [9]. Imaging makes it possible to make the diagnosis to assess the maturation of the lesions and to seek the associated locations. Bone scintigraphy can be used for diagnosis and for follow-up; it is the most sensitive imaging test for detecting POA [9]. It is usually positive after 2 to 4 weeks from the start of the bone formation process, and also makes it possible to assess the maturity of POA. Many teams have shown that waiting for POA maturity is no longer justified in considering its exeresis, this examination has therefore become exempt. Standard radiography, MRI and CT have low specificity at the initial stage [6,11]. Ultrasound can detect POA earlier than with conventional radiographs [12]. Local signs of inflammation in the spinal cord injury are in favor



Figure 4: Pelvis X-Ray after one year of monitoring.

of POAN [13]. It is undoubtedly the best examination, not only for early detection, but also for monitoring POA [9]. Its sensitivity in the early phase to diagnose POAN in the BM patient has been estimated at 89% [14]; it is thus carried out in certain centers on a bi-weekly basis for early systematic screening [14]. The CT scan makes it possible to demonstrate bone formation after 3 weeks, while standard radiographs only reveal the osteoma after 4 to 6 weeks [15]. Although the latter are sufficient to highlight the annoying POA, the scanner is the examination of choice to assess the operational risks because it provides information concerning the bone density of the joint underlying the POAN [16], but also on the relationships between POAN and adjacent structures, after 3-dimensional reconstruction. The injection of contrast medium will allow good visualization of the vessels. All of these elements will allow the surgeon to choose a pathway for excision of the POAN [16].

Even today, the only validated and effective treatment remains surgery to remove heterotopic ossifications [8,9]. It improves joint mobility [6,17] as well as passive and active function [6], reduces pain [6] (by nervous decompression) and improves the development of pressure sores. It has long been thought that resection of POANs should not be done before they have matured. However, no adequate randomized controlled study has been performed to confirm this principle and resection of immature heterotopic ossification does not predict a higher rate of recurrence [18,19]. The delay in relation to the accident does not seem to be a determining factor since the maturity of the POANs does not influence recidivism [8,9,17]. In addition, an extended surgical delay leads to ankylosis which is of poor prognosis for functional recovery. The excision surgery can be performed in patients with extensive neurological impairment as soon as comorbidities are under control [9,20], even in patients with a major neurological handicap which is linked to their initial impairment. The CT scan is essential to determine the bone mineral density as well as the intra articular lesions. In addition, thanks to 3-dimensional reconstructions, it can be used to assess the proximity of the osteoma to neurovascular structures and thus guide the choice of surgical approach [20]. Genêt et al. have studied the impact of late surgery on hip POAN after neurological injury of traumatic origin [20]. They highlighted that the loss of mobility before ankylosis was a more important factor than the maturity of the latter in deciding when to intervene. Early intervention minimizes invasion and intra-articular damage, demineralization, as well as post-surgical complications such as per or postoperative fractures [17,20] without increasing the risk of recurrence. Among the 24 patients who underwent elbow POAN surgery, Lazarus et al. have shown that long surgical delays

are a negative predictor of recovery in joint mobility [21]. Surgical resection of the heterotopic ossification is followed by prophylactic measures, given its tendency to recur.

Conclusion

Neurogenic para-osteo-arthropathies are classic complications of central neurological conditions, especially in traumatic contexts. They occur mainly in the vicinity of large joints. Their exact pathophysiology remains unknown despite numerous studies and reported cases. Surgery retains its essential place in their management despite the recurrence rate estimated between 17% and 58% [22].

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