SPMRJ Volume 6 No. 2 August 2024 DOI: 10.20473/spmrj.v6i2.48341
SINTA 5 (Decree No: 200/M/KPT/2020) Available online at https://e-journal.unair.ac.id/SPMRJ

Case Report
Neurogenic Arthropathies of the Lower Limbs: About A Case Followed in Rehabilitation at the Chu-Bogodogo

Tiaho Y[1], Sougué C[2], Tougma G-WL[1], Traoré S.I[3], Sebgo B[1], Bamogo B[1], Gandema S[4].
1. Department of Physical and Rehabilitation Medicine , University Hospital Bogodogo
2. Department of Rheumatology, University of Hospital Bogodogo
3. Department of Anesthesia and Intensive Care, Bogodogo University Hospital
4. Departement of Physical and Rehabilitation Medicine, University Hospital Souro Sanou

Correspondent:
TIAHO Yirozounlomian
Joseph KI-ZERBO University (UJKZ),department of health sciences
Ouagadougou-Burkina Faso
E-mail: yiro.tiaho@ujkz.bf

Article info:
Received: August 8, 2023
Received in revised: May 15, 2024
Accepted: June 20, 2024
Published: August 28, 2024

Cite this as: Tiaho Y, Charles S, Louis T, S.I T, Boukari S, B B, Salif G Neurogenic Arthropathies of the Lower Limbs: About A Case Followed in Rehabilitation at the Chu-Bogodogo. SPMRJ Vol. 6 No. 2, 159-173

Abstract

Traumatic brain injuries can be worsen during hospitalization by pelvic limb neurogenic arthropathies (NA). The NA is responsible for painful periarticular and muscular inflammatory phenomena, which develop into partial ossification of the periarticular soft structures. This partial ossification will be responsible for muscular and articular retractions limiting walking. The purpose of the report was to demonstrate the contribution of cryotherapy and joint mobilization techniques in limiting the progression of NA. The patient was 25 years old and was admitted to the physical medicine and rehabilitation department 6 months after a severe head injury. Nervous system disorders were represented by a frontal syndrome and significant spasticity in the pelvic limbs. Deficiencies found were pain, multiple joint stiffness of the pelvic limbs, and induration of the thighs related to arthropathies objectified to the standard x-ray of the thighs. Rehabilitation consisted of posture sessions on a standing table and cryotherapy at the end of the session. The gait rehabilitation was done gradually between the parallel bars and then with a deanbulator. Rehabilitation led to functional improvement, with the patient returning to independent walking using English walking sticks. The presentation of this case study provided an overview of this pathology, its functional consequences, and the role of physical and rehabilitation medicine in the management of NA. The techniques we used helped to reduce the NA by limiting inflammation with cryotherapy sessions and recovering joint amplitude with progressive posture techniques. The presence of nervous system disorder such as frontal syndrome could have limited rehabilitation, but thanks to collaboration with psychiatrists, the evolution was favorable and allowed the patient to actively participate in his rehabilitation. The case also underlined the importance of multidisciplinary patient management, which should be a goal for all practitioners.

Keywords: *arthropathies; cryotherapy; neurogenic; rehabilitation; traumatic brain injury*

INTRODUCTION

Neurogenic arthropathies (NA) are abnormal formations of mature lamellar bone in extraskeletal soft tissues, like muscles, tendons, and ligaments. They develop mainly in the periarticular soft tissues, frequently following a central neurological lesion, most often traumatic.[1-6] NA occurs in 10-20% of head trauma patients.[7,8] This process is thought to be mediated by inflammation associated with tissue damage.

Treatment with nonsteroidal anti-inflammatory drugs and low-dose localized irradiation are currently the only prophylactic treatments available for NA. However, they are not always effective and do not directly target osteogenic processes. New therapeutic strategies targeting the pathological processes of NA, such as bone morphogenetic protein (BMP) inhibitors like noggin, BMP type 1 receptor inhibitors, and nuclear retinoid-gamma acid receptor (RARγ) agonists, are currently being investigated. The curative treatment of NA is based on surgical treatment by excision, which is often complicated by recurrence.[9,10]

None of these methods is currently the subject of a specific recommendation. However, the literature seems to indicate that the treatment of NA is based on the resection of the ossification, with adjuvant measures such as nonsteroidal anti-inflammatory drugs, bisphosphonates, radiotherapy, and physical therapy.[11]

Like its poorly codified treatment, NA is the cause of limitations in joint mobility and pain that hinder the positioning and function of the musculoskeletal system. Thus, the occurrence of NA in the pelvic limbs can have serious repercussions on transfers, walking, and a significant limitation in the activities of daily life.

In the literature review, rehabilitation was not cited as a means of preventing or treating NA. In any case, the medicinal treatments administered during NA are aimed at limiting inflammation of the soft tissue and delaying ossification. Cryotherapy is used to combat inflammatory phenomena and is widely used in rehabilitation settings. In addition, in the case of joint and muscle limitations linked to NA, surgery has been indicated, but with

frequent recurrences. In view of the pathophysiology we now know, cryotherapy and rehabilitation could be used to prevent and treat NA. Joint and functional limitations could also be treated through rehabilitation.

The objective of this report is to describe a case of heterotopic ossification in the pelvic limbs after a head trauma, presenting the clinical manifestations and discussing the treatment instituted by physical methods that allowed a good functional recovery in a young patient who developed NA of the pelvic limbs following a serious head trauma caused by a road traffic accident.

CASE REPORT

The patient was 25 years old, with no known pathological history. He was hospitalized in the intensive care unit on 22 August 2022, following a serious cranio-encephalic trauma with a coma from the outset that occurred in a road traffic accident. The accident occurred on a motorcycle, and the patient was not wearing a protective helmet. Our patient benefited from resuscitation associated with nursing without maintenance of joint mobility due to a lack of rehabilitation personnel.

The evolution was marked by an improvement in the state of consciousness and his transfer to a neurosurgery department 1 month after his hospitalization in intensive care. Additional explorations carried out in neurosurgery will reveal the presence of an acute left hemispheric subarachnoid hematoma and an acute right sub-dural hematoma associated with COVID-19 pneumonia, which will be successfully treated.

The clinical evolution was then marked during hospitalization in neurosurgery on November 1st by the progressive appearance of painful swellings of the thighs, which motivated the realization of an ultrasound of the soft parts, which revealed a fluid collection periosseous femoral and intra-articular of the knees with the beginning of ossification, fusing in the intermuscular of the quadriceps until the proximal 1/3 of the thighs (image 1). Given the impossibility of performing a CT scan for financial reasons and the availability of the technical platform, standard thigh x-rays were requested in

addition. These radiographs made it possible to objectify intramuscular and periarticular ossification processes in both thighs (image 2, image 3, image 4). The patient will be discharged from the neurosurgery department at the end of November 2022 with a prescription for tablet tramadol for pain and physiotherapy sessions. Physiotherapy sessions were carried out at home at the parents' request. These home sessions consisted of turning the patient over in bed, transfers, and placing the patient in a wheelchair. Mobilizing the joints was painful, and standing was impossible, even with help. Because he was unable to walk and the stiffness and pain persisted until February 2023, the patient was referred to the physical medicine and rehabilitation department of the Bogodogo University Hospital (CHU-B). We received the patient on 23 February 2023 and his admission report noted:

- General and segmental mobilization pain rated on the visual analog scale (VAS) at 6/10;
- A frontal syndrome (disinhibition, perseveration, stereotyped behaviors, attention disorders, praxis disorders);
- Joint stiffness involving the large joints of both pelvic limbs (image 5);
- Ankle in bilateral equinus of 30° and knees at 50° in active and passive;
- A motor deficit (paraparesis: 3+ proximal and 3- distal to the 4 limbs);

Spasticity affected the pelvic limbs and was rated at 3/5 on the modified Ashworth scale, with bilateral equinus of the feet, and the patient had several activity limitations rated according to the functional independence measure (FIM)[12] at 18, which corresponds to total assistance in the ADLs for more than 8 hours.

We have set up a rehabilitation protocol of 4 sessions per week on an outpatient basis, lasting 1 hour, and under analgesics to fight against pain, which included extension postures of the pelvic limbs using straps and standing on a table combined with infrared therapy for circulatory and analgesic purposes during the posture phases (image 6) and cryotherapy with ice at the end of the session; then, work on walking between parallel bars and with a walker (image 7).

The drug treatment included:

- Tramadol tablet 50 mg for pain (1 tablet morning and evening and 1 tablet 1 hour before the sessions;
- Piroxicam 20mg capsule (1 capsule per day to limit the progression of NAOPs and fight against pain);
- Lioresal 10 mg tablet for spasticity at progressive doses up to 3 tablets/day (1 tablet morning, noon and evening);
- Largactil 100mg tablet on the advice of the psychiatrist (1/2 tablet morning and evening).

Weekly clinical monitoring (pain, behavioral problems, motor deficit) and monthly biological monitoring (creatinine, ASAT/ALAT, NFS) were instituted.

The evolution was marked by a gain in joint amplitude in the knees and ankles without any pain; and having allowed walking with technical assistance (image 8) and an improvement in the FIM score, which rose to 90, corresponding to ADLs supervision over a time of less than 2 hours. There was also a satisfactory improvement in spasticity and frontal syndrome.

DISCUSSION

This clinical observation recalls once again the problem of cranio-encephalic trauma from a traffic accident. These traumas affect much more young adult males, who seem to be more exposed, and in whom the severity of these injuries in developing countries such as Burkina Faso is closely linked to the non-wearing of helmets among these young motorcyclists.[13] These are the same circumstances of occurrence according to the history of the disease of our patient, who was not wearing a helmet at the time of the accident with his motorcycle. It should be noted that in Burkina Faso, whose capital city of Ouagadougou is known as the capital of two-wheeled machines, users do not wear protective helmets. This situation calls into question the awareness of the wearing of helmets and the application of the law on the non-wearing of helmets in our developing countries. This will reduce the velocity of shocks at the time of accidents and preserve the vital and functional prognosis in these

motorcycle accident victims. In addition, our patient's COVID-19 test came back positive during hospitalization. Cases of NA were described during the COVID-19 pandemic;[14] this means that in addition to the head injury, the patient had another risk factor for developing NA due to his Covid-19 infection. Our patient was treated in intensive care because of his coma and the vital prognosis, which was at stake; however, the preservation of the functional prognosis regardless of the state of the organic lesions presented by our patient was not taken into account during his stay in intensive care. The management of patients in intensive care should take into account the prevention of complications of immobility and decubitus, including joint and muscle maintenance, which requires gentle physiotherapy at this phase. In our context of Burkina Faso, physical medicine and rehabilitation remains embryonic, and the country has no complete hospitalization service for rehabilitation care suites.

To this, it is necessary to note the insufficiency of rehabilitation personnel, in particular physiotherapists. In Burkina Faso, public rehabilitation facilities are operational in five university hospital centers (CHU) and two regional hospital centers (CHR). There are a total of 42 private facilities spread over 22 provinces, with a high concentration in Ouagadougou, the country's capital. However, the existing infrastructure is not adapted (access ramp, size of openings, position of toilets), and the premises are often cramped and dilapidated. With regard to PRM materials and equipment, there is a lack of availability due to very high costs and a poorly developed supply system. Most equipment is imported. In addition, the human resources of the various PRM professions are very limited (6 PRM specialists, 60 physiotherapists, including 4 at higher levels, and 19 orthoprosthetists). This has resulted in a low supply of care, poor quality of care, and poor patient safety in PRM. This situation means that rehabilitation care is not systematically included in the acute phase during hospitalizations of head trauma patients. If NA seems to be addressed in the literature, in practice, their prevention, diagnosis, and management remain little known to many practitioners. Rehabilitation is not

mentioned in the literature as a means of managing NA. The pathophysiology of NA suggests an inflammatory process, which justifies the use of anti-inflammatory drugs for treatment and prevention. Rehabilitation offers a number of techniques for combating inflammation, such as cryotherapy. The inflammatory phenomenon caused by NA leads to progressive stiffening of joints and muscular structures, which ossify and become inextensible. Gentle mobilization by kinesitherapy is highly beneficial in preventing joint stiffness and, above all, maintaining supple muscle structures. Physiotherapy should be started as early as possible, even in the acute phase. There are no national or even international recommendations on the management of NA. This pathological process could go unnoticed by practitioners, while its functional impact can be very important, with stiffness, pain, and functional impotence affecting the patient in the aftermath of hospitalization. Rehabilitation is indicated to preserve joint mobility and, above all, to combat the inflammatory phenomena associated with NA. In our case, the patient was seen with many joint limitations and ossification processes in both thighs. Bed transfers, standing and walking were impossible for our patient. As a result, he was severely limited in his activities of daily living. In the literature, risk factors such as the occurrence of head trauma or surgery have been listed.[6,8] In the case of our patient, the pathological process would have appeared 1 month after his trauma by the progressive appearance of painful swellings on both thighs. Our patient also tested positive for COVID-19 during his stay in intensive care. This COVID-19 infection would certainly be linked to a nosocomial etiology. COVID-19 has also been suggested as a risk factor in some studies. It is certainly understandable that COVID-19 should be evoked as a risk factor, given that the pathophysiology of COVID-19 infection involves a major inflammatory process affecting not only the lungs but also the blood system, leading to pulmonary embolism in patients. This inflammatory process affecting the blood could reach the muscular system by diffusion during infections, in particular that of COVID-19, which is responsible for a significant

inflammatory response. According to the literature, treatment with an anti-inflammatory would have made it possible to reduce the process and thus prevent the heterotopic ossifications (15) found on radiology and ultrasound. Current data have identified celecoxib, naproxen and diclofenac as the best option for the prevention of NA in patients undergoing primary arthroplasty. The most appropriate pharmacotherapy for the prevention of NAOPs is still debated and needs to be personalized according to patients' comorbidities and medical history. For patients with cardiovascular comorbidities, naproxen or diclofenac should be considered alongside proton pump inhibitors to prevent gastrointestinal complications. For patients with a history of gastrointestinal disease, celecoxib may be recommended. Apart from anti-inflammatories, we have also palovarotene (Sohonos™) which is an orally bioavailable, selective retinoic acid receptor (RAR)γ agonist developed by Ipsen to reduce heterotopic ossification (HO) formation in patients with fibrodysplasia ossificans progressiva (FOP). By binding to RARγ, palovarotene inhibits bone morphogenetic protein and SMAD 1/5/8 signaling: interference with these pathways

prevents chondrogenesis and, ultimately, heterotopic ossification by allowing repair or regeneration of normal muscle tissue. Palovarotene received its first approval on January 21, 2022 to reduce HO formation in adults and children aged 8 and over for women and 10 and over for men with FOP in Canada.[16] Although in our patient this anti-inflammatory treatment was not instituted because of their non-safety; a method of gentle physiotherapy and cryotherapy would certainly have made it possible to reduce the inflammatory phenomenon observed during the pathological process of NA. A collaboration between resuscitation, neurosurgery and PMR would have made it possible to establish the diagnosis and to institute an adequate management protocol on multidisciplinary consultations. The onset of thigh pain in hospitalized patients with risk factors such as head trauma, surgery or infection, particularly with COVID-19, should prompt practitioners to look for NAs. The diagnosis can be confirmed by soft tissue ultrasound or magnetic resonance imaging. Once the diagnosis has been confirmed, and if there is a contraindication to the use of anti-inflammatory drugs, gentle

physiotherapy and cryotherapy should be considered, as these techniques are highly effective in combating the inflammatory phenomenon and joint stiffening. In fact, the physiological explanation for the effect of cryotherapy has already been demonstrated in an animal model:[17,18] the effects of cryotherapy combined with therapeutic ultrasound on oxidative stress and tissue damage after musculoskeletal contusion in rats showed excellent results. In fact, cryotherapy effectively combats the inflammatory process by reducing the oxidative stress caused in injured tissue.

Ultimately, early involvement of the PMR would have prevented NA in this patient. In addition, the absence of a complete hospitalization unit in our country led the patient's family to decide on home physiotherapy. This phenomenon is observed a lot in our context because for patients with reduced mobility, there is no adequate transport mechanism and the cost of transport is twice the price of a single session. A study of access to care for vascular hemiplegic patients in the city of Ouagadougou found that the average cost of a rehabilitation session was 5953.12 +/- 1899.25 FCFA (about 9 euros) for the private sector and 3377.60 +/- 564.22 (about 5.5 euros) for the public; that of transport was 2656.25 +/- 1574.53 FCFA (about 4 euros) for personal transport and 3343.75 +/- 491.13 (about 5 euros) for public transport.[19] this situation leads patients to decide in favour of home rehabilitation sessions. This decision raises the issue of home physiotherapy care for patients with very reduced autonomy and presenting with multiple stiffnesses. Manual postures can be very limited and the absence of a standing table and other rehabilitation materials means that home sessions should not be indicated for very dependent patients, in whom the functional benefit of rehabilitation is still present. In our patient's case, adapted and progressive postures on the standing table enabled joint amplitudes to be recovered. The infrared physiotherapy technique was used to prepare the joints for the various postures, and cryotherapy was used at the end of the sessions for its analgesic and anti-inflammatory effect. The rest of the re-education consisted of progressive transfers to bed and chair. The

use of parallel bars enabled the patient to stand upright, and to begin progressive work on walking and proprioception. Drug treatment with largatil helped to overcome frontal syndrome and encourage the patient's own involvement in his rehabilitation. This quickly produced satisfactory functional results. Although spasticity was a problem from the outset for active joint mobilization and walking, the administration of lioresal to our patient enabled him to combat this spasticity.

Our patient was able to benefit from comprehensive care in the PMR department with an outpatient rehabilitation protocol only six months after his cranioencephalic trauma; with functional improvement in our patient and good autonomy in activities of daily living. However, this delay could seem long because beyond the vital prognosis, the practitioner should always think about the functional prognosis in patients in acute hospitalization. The organization of care in Africa, in the absence of rehabilitation care units, should encourage the collaboration of acute care services with PMR services. This calls for recommendations on the diagnosis of NA, the identification of risk factors for

NA and existing therapeutic modalities, taking into account physiotherapy techniques as an alternative to anti-inflammatory drugs.

CONCLUSION

This clinical observation reminds the practitioner of the occurrence of NA in a certain number of clinical circumstances, in particular during head trauma. Their functional impact can be serious and therefore, adequate prevention and the prescription of rehabilitation care in the acute phase could help prevent the occurrence of NA or, failing that, improve the functional prognosis in head trauma patients.

DISCLOSURES

Acknowledgment

We would like to thankMr CISSE, a teacher at Joseph KI-ZERBO University, for his invaluable help in translating this document.

Conflict of interest

The authors declare that they have no conflicts of interest.

Funding

This study has not received any funding.

Author Contribution

Study design and plan: Tiaho Yirozounlomian and Sougué Charles. Data

collection: Tiaho Yirozounlomian, Sebgo Boukari and Bamogo Boureima. Data analysis,interpretation and drafting of manuscript: Tiaho Yirozounlomian and Tougma Louis. Revision of manuscript: Tiaho Yirozounlomian and Gandema Salifou. Guarantor for the study: Tiaho Yirozounlomian

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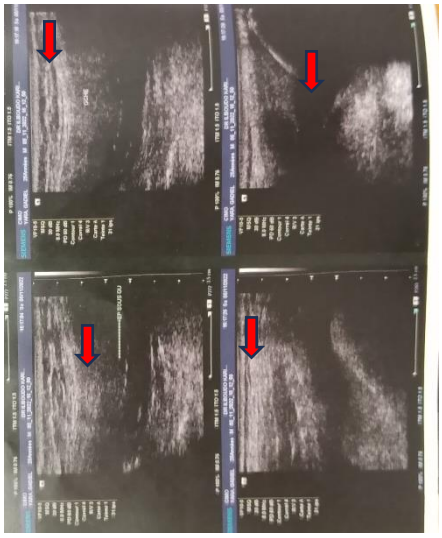

Image 1: Ultrasound of soft tissue showing periosteal fluid collection in femur and intra-articular knee with ossification.

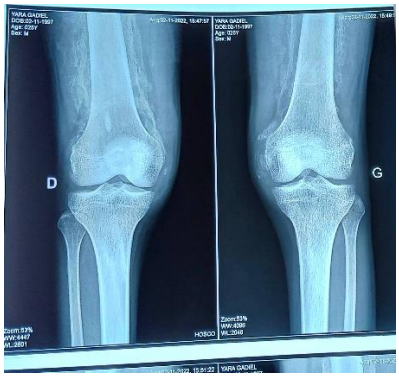

Image 2: frontal X-ray of the knees showing ossifications in the soft tissues of both thighs

Image 3: Profile X-ray of the knees showing ossifications in the soft tissues of both thighs

Image 4: Ossification along the thigh

Image 5: Spontaneous positioning of the patient in bed, with the right pelvic limb in external rotation, knee flexed and equinus foot resting on the lateral edge.

Image 6: Prone posture for amplitude recovery in extension

Image 7: walking between parallel bars

Image 8: walking independently with a walker