Effects of Aquatic Physical Therapy in Children With Congenital Femoral Deficiency Who Underwent Bone Lengthening Using an External Fixator: A Case Report

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Introduction and Objective: Congenital femoral deficiency (CFD) is a malformation that affects 0.2 persons per 10,000 live births. The orthopedic treatment proposed for this disease is bone lengthening using an external fixator (EF). During this treatment, reductions in the range of motion (ROM) of the knee, alterations in the gait speed (GS), and worse quality of life (QOL) commonly occur. Therefore, rehabilitation of patients with CFD receiving this treatment is essential. Aquatic physical therapy (APT) is an innovative approach for this population, however, there are no reports on the effects of APT in patients with CFD after bone lengthening. The objective of this study was to assess the influence of APT on the knee ROM, GS, and QOL of 2 children with CFD implanted with an EF.

Case Description: Two children with right-sided CFD implanted with an EF were selected. They received an APT protocol focusing on balance, muscle activation, analgesia and mobility of the right knee, and gait. The right knee ROM, GS, and QOL were measured using right knee goniometry, a 10-m walk test before and after each session, and the Child Health Questionnaire–Parent Form 50 before and after the protocol.

Results: An increase in the ROM of the right knee and an improvement in the QOL were observed after the protocol. Only one of the children had an increased GS.

Conclusion: The APT protocol increased the knee ROM and improved QOL of the 2 children with CFD implanted with an EF. (J Aquat Phys Ther 2022;30(3):65–69)

Key words: bone lengthening, congenital femoral deficiency, hydrotherapy

Congenital femoral deficiency (CFD) is a rare malformation of the femur of unknown cause that can range from hypoplasia to the total absence of the femur. This alteration occurs in the embryonic period, from the 4th to 8th weeks of intrauterine life, causing dysfunction of the hip and knee joints. Its incidence is 0.11 to 0.20 per 10 000 births, and 85% to 90% of cases have unilateral involvement.[1]

Several classifications have been described to guide the treatment and improve the prognosis using radiological examinations. Paley's classification is the most commonly used classification system, which considers several factors that influence the reconstruction of the femur.[2] This classification has 4 types and subdivisions, which describe the distal femoral malformation from preserved and progressing knee mobility to the complete absence of the femur.[2]

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Typically, the treatment of CFD consists of femoral lengthening with the use of either an external fixator (EF) or a magnetically motorized intramedullary rod.[3] During the bone lengthening process, it is common for patients who have a present knee joint to have a decreased range of motion (ROM) of the knee, resulting from inflammation and pain in the region where the pins are attached, as well as from muscle atrophy and loss of strength, a balance deficit, and asymmetries in gait.[4]

The objective of physical therapeutic rehabilitation is to promote independence and ambulation. It is important to maintain or improve the functional knee ROM, increase muscle strength and weight-bearing capacity for a smoother and more controlled gait pattern, improve balance, and prepare the patient for activities of daily living.[5]

Aquatic physical therapy (APT) is a possible intervention for patients with CFD because it helps reduce pain and edema, decreases joint overload, increases the degrees of freedom of movements, facilitates safe gait training, stimulates proprioception, and increases muscle activation.[6,7] These benefits are promoted by the physical and thermodynamic principles of water, which can facilitate or hinder any type of movement.[7] Hydrostatic pressure aids venous return and the transport of fluids that are important factors of both analgesia and inflammation, along with temperature.[8] The level of immersion at the height of the xiphoid process, along with the thrust, decreases

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the percentage of weight-bearing on the joints, facilitating anti-gravitational movements, such as knee flexion and hip flexion.[9]

It is known that mineral deposition in the bone structure stimulates consolidation.[10] The reduction of the weight load while in the water occurs owing to the thrust action; however, it has been observed that this facilitates muscle activation because the force exerted by the muscle that contracts against a resistance produces a mechanical stimulus in the bone, and this stimulus is similar to that resulting from a weight load. Weight-bearing can also be facilitated by the viscosity of water, which allows the individual to perform activities with greater safety, since it promotes an increase in the response time to imbalance,[8] contributing to exercises being performed with unilateral support. Turbulence can be used to promote analgesia, in addition to providing some balance instability. The physiological effects and scientific evidence of the effectiveness of APT influence the domains of quality of life (QOL).[11,12]

Considering the benefits of water on an immersed body and the problems observed in individuals with CFD implanted with an EF, the objective of this study was to verify the effects of APT on the knee ROM, gait speed (GS), and parents' perception of the QOL of 2 children with CFD implanted with an EF.

METHODS

This case study involved 2 patients, a 10-year-old boy and an 8-year-old girl, who started femoral bone lengthening treatment using a unilateral EF less than 6 months ago. Both patients had DCF type 1a according to Paley's classification and a 4-cm discrepancy in the right lower limb. They were in the consolidation phase, with the EF implanted in the right femur. The patients followed an APT exercise protocol for 4 weeks, with 35-minute sessions once a week, with the water temperature at 33 °C. They also underwent conventional physical therapy once a week, on different days from APT, for 35 minutes, with exercises focusing on body weight unloading and muscle strengthening. The evaluations were performed before and after each APT intervention, which made it possible to measure the results of each APT session in isolation.

During APT, it is important to avoid trauma and mechanical shock to the affected limb. The patient and their companion must be instructed to wash the pin incision area with running water and perform adequate hygiene after every therapy session. In the case of signs of inflammation or infection in the pin incision, liquid medium therapies must be suspended, and a medical evaluation will be necessary. All these precautions were taken before placing the patients in the aquatic environment for therapy. No complications occurred at any time, and the patients had periodic medical follow-up.

Case Description

The 2 children underwent 3 evaluation procedures. The ROM of the right knee and the GS were evaluated before and after each intervention, and the parents' perception of their children's QOL was evaluated before and after the APT protocol.

Goniometry[13] of the flexion and extension of the knee was used to measure the ROM of the right knee, starting at the maximum extension amplitude of the right knee, with the patient in dorsal decubitus, positioned at 90° of hip flexion, and with the fixed arm of the goniometer parallel to the greater trochanter, the axis of the goniometer parallel to the knee joint, and the mobile arm of the goniometer parallel to the tibia.

To evaluate the GS, we used the 10-m walk test.[14] For this test, the patients were initially in the standing position, with the support of bilateral forearm crutches and with shoe compensation of the affected lower limb. At the evaluator's command, the patients walked for 10 m, having been previously instructed to walk as fast as possible safely and without running. The evaluator used a digital stopwatch to count the time.

Finally, the Child Health Questionnaire–Parent Form 50[15] (CHQ-PF50) was administered to assess the parents' perception of their children's QOL. This is a generic instrument for assessing health-related QOL in children older than 5 years and adolescents, aiming to determine the patients' physical, emotional, and social well-being from the perspective of their parents or guardians. The questionnaire was administered by the evaluator before the first session and after the last session of the APT protocol.

The intervention protocol in the aquatic environment consisted of the following:

- Static passive stretching of the ischiotibial muscles of the affected limb for 60 seconds in the sitting position while immersed in water;
- Static passive stretching of the knee extensors of the affected limb for 60 seconds in the sitting position while immersed in water;
- 10 minutes of exercises simulating pedaling, in the vertical position and supported by floats, while immersed up to the level of the xiphoid process;
- Unipedal support of the affected lower limb in the standing position 5 minutes, with the contralateral lower limb placed on a step while immersed up to the level of the xiphoid process and with turbulence generated in the direction of the affected limb;
- Picking up of 10 rings with the foot of the affected lower limb, in the standing position, while immersed up to the level of the xiphoid process, repeated 3 times;
- Three sets of 20 repetitions of active flexion of the knee of the affected lower limb, in the standing position, while immersed up to the level of the xiphoid process; and
- Independent march for 5 minutes while immersed up to the level of the xiphoid process.

Figure 1 shows one of the patients implanted with an EF performing an activity in the pool.

Data Analysis

The data were computed using Microsoft Excel. A descriptive analysis between the data on the knee ROM and GS obtained before and after the intervention in each session was performed. Data on the QOL were analyzed before the first session and

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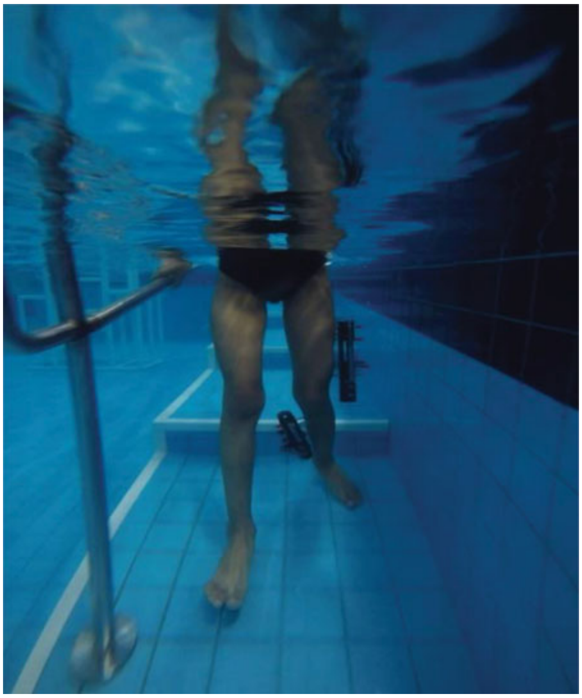

Fig. 1. Patient implanted with an external fixator entering the pool to undergo aquatic physical therapy.

after the last session of the protocol. The results were described individually for each of the cases analyzed.

RESULTS

An increase in the ROM of the direct knee was observed after each APT intervention and after the entire protocol established for the 2 children. Figures 2 and 3 show the male and female patients' ROM progression throughout the sessions, respectively.

The male patient showed an increased GS after each intervention and at the end of the protocol. The female patient did not show an increase in the GS after any session or at the end of the protocol. Figures 4 and 5 show the male and female patients' GS progression over the APT protocol, respectively.

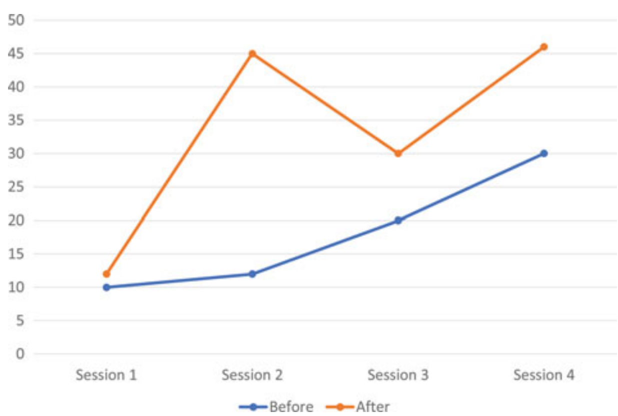

Fig. 2. Range-of-motion progression of the right knee of the male patient (presented in degrees).

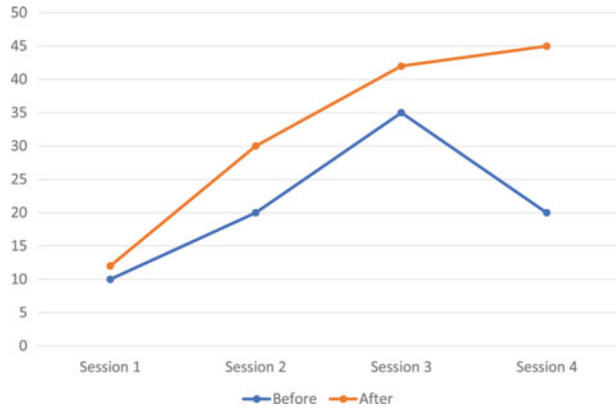

Fig. 3. Range-of-motion progression of the right knee of the female patient (presented in degrees).

Both patients showed improvements in most QOL domains in the CHQ-PF50. Figures 6 and 7 show the changes in the QOL domains after each session and before and after the established protocol in the male and female patients, respectively.

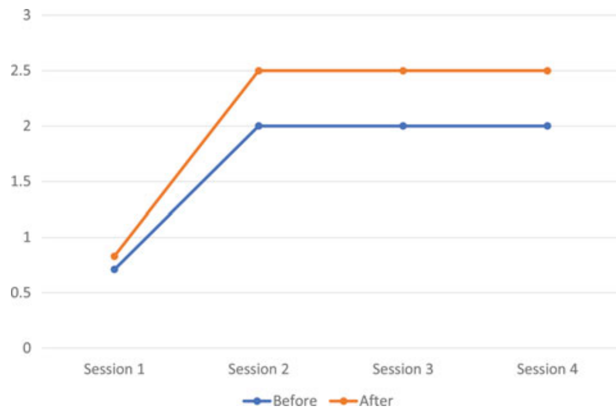

Fig. 4. Gait speed progression of the male patient (presented in meters per second).

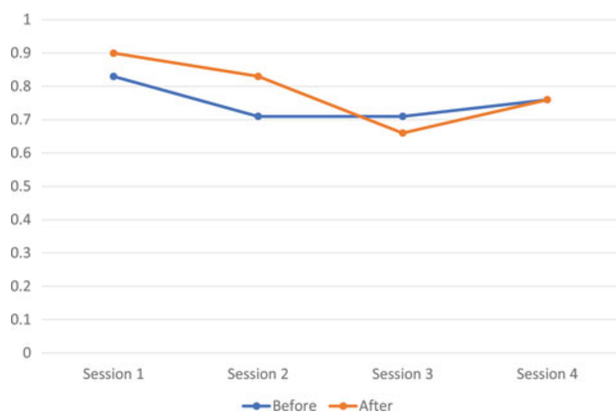

Fig. 5. Gait speed progression of the female patient (presented in meters per second).

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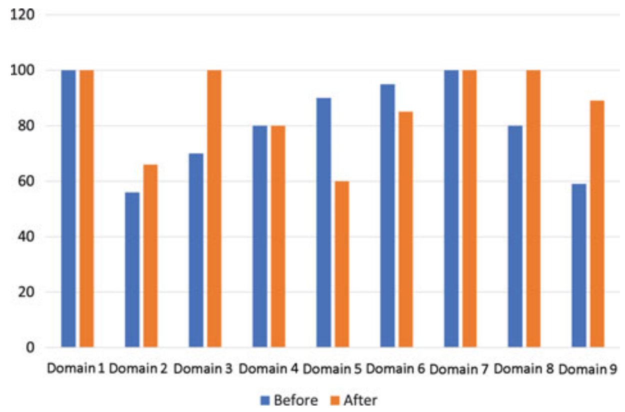

Fig. 6. Child Health Questionnaire–Parent Form 50 scores before and after the intervention in the male patient.

DISCUSSION

APT can aid in achieving functional gains for patients implanted with an EF for bone lengthening. However, APT is rarely an object of research. No standardized procedures have been described, and there are few reports of procedures to date.[16] During the consolidation phase of femoral lengthening, the knee ROM, as well as muscle strength, should be maintained as functional as possible to ensure treatment success.[5]

In this study, the patients initially demonstrated a decrease in the knee ROM of the affected limb owing to the fixation of the pins in the musculature, which impacts the knee joint's capacity for adequate movement. This is a very frequent finding in this population.[3] After the APT intervention period, there was an increase in the knee ROM, an increase in passive flexion, and a decrease in residual flexion at rest. These gains were noticed immediately after the intervention and throughout the sessions.

We believe that these gains are the result of static stretching and active exercises, such as pedaling, active knee flexion in the standing position, and knee flexion when picking up rings with the foot of the affected limb. The execution of these exercises was directly related to the physical and thermodynamic principles on the body in immersion. Warm water promotes muscle relaxation, which facilitates the stretching and mobilization of the joints and body segments with the knee flexors and extensors.[9]

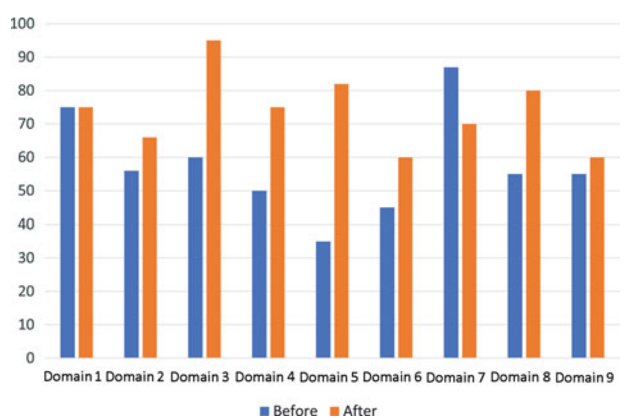

Fig. 7. Child Health Questionnaire–Parent Form 50 scores before and after the intervention in the female patient.

An increase in the GS was observed in the male patient after the interventions, demonstrating functional improvement and the positive transference of the gains obtained in each APT session. This did not occur in the female patient, whose GS decreased. The gait training performed in the pool was included in the protocol to promote the experience of the activity and the subsequent repercussion on the ground after the protocol. The GS may not be a sensitive parameter to score this population's functional improvement. An evaluation using movement analysis instruments may be necessary to evaluate the spatial-temporal parameters and the asymmetries of gait, observing the affected limb in a more global manner.

The QOL of the male patient, as perceived by his parents, improved in 7 of the 9 CHQ-PF50 domains. The "well-being" and "behavior" domains were the only domains for which the patient's mother did not notice any improvement. Meanwhile, the QOL of the female patient, as perceived by her mother, improved in all domains, except for "well-being." We believe that the children implanted with an EF were dissatisfied from an aesthetic point of view, and after the proposed protocol started, the sequence of activities may have interfered in the routine of their families, impacting the perception of well-being and behavior. The aquatic environment is not only motivating but also challenging. These aspects may have influenced the well-being of the patients because it is known that repetitive exercise protocols can become tiring and demotivating; motivated participants are known to contribute to a better therapeutic response.[17] Another study observed that in addition to motor improvement, an aquatic activity program yielded benefits in social function, which consequently influenced the QOL.[11,12]

As mentioned previously, warm water promotes analgesia, a fact that may explain the increased score in the "decrease in pain" domain of the CHQ-PF50. The literature reinforces the idea that one of the main effects of immersing the body is promoting analgesia. In addition to the effects of temperature, a sensory overflow occurs in the immersed body, which reduces the perception of pain in the affected limb. It has been suggested that just by the child being in contact with the liquid medium, there is already a reduction in the perception of pain.[18]

The use of a liquid medium in the rehabilitation process during the use of an EF is still very rare. Patients who receive this type of intervention rarely have access to a rehabilitation center and to trained professionals (orthopedists, rehabilitation physicians, and physical therapists) who offer this service. Some studies[5,9] report that patients implanted with an EF who undergo APT after hospital discharge need care to prevent complications and infections. The incision area of the pins should be clean. After every session, the patient and their caregiver should wash the pin incision area with running water and perform adequate sanitation. In the case of signs of inflammation or infection in the pin incision site, liquid environment therapy must be suspended and a medical evaluation will be required. Herein, all these precautions were taken; no complications occurred, and the patients had periodic medical follow-up.

During this study, the patients had already completed the bone lengthening phase and were in the phase of bone healing, which allowed adequate and early weight load throughout

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the prescribed exercises. They demonstrated no complications during the intervention protocol.

There is a scarcity of literature related to the rehabilitation of patients with CFD implanted with an EF, particularly to rehabilitation in an aquatic environment. In this study, an increase in the ROM of the right knee was observed in both patients after all APT sessions, in addition to an improvement in the QOL, as perceived by their parents, after the protocol. This study demonstrates that APT promotes positive effects in patients with CFD implanted with an EF.

This case study had a small sample size, which limits the generalization that the APT protocol can be extended to the entire population with CFD implanted with an EF.

WHAT DOES THIS CASE ADD TO EVIDENCE-BASED PRACTICE?

In this study, it was observed that functional APT promoted an increase in the knee ROM and an improvement in the parents' perception of the QOL of 2 children with CFD implanted with an EF for bone lengthening. This study may serve as a reference for future studies and possible clinical trials with larger samples to either support or refute the findings. The use of APT in this population has not been well addressed in the previous literature and is scarcely studied but is extremely important for the rehabilitation of these patients.

REFERENCES

1. Valentina D, Gaia P, Antonia S, et al. Prenatal diagnosis of proximal focal femoral deficiency: literature review of prenatal sonographic findings. *J Clin Ultrasound*. 2016;44(4):252-259. doi:10.1002/jcu.22306
2. Rozbruch SR, Ilizarov S. *Limb Lengthening and Reconstruction Surgery*. Inform Healthcare; 2007.
3. Szymczuk VL, Hammouda AI, Gesheff MG, Standard SC, Herzenberg JE. Lengthening with monolateral external fixation versus magnetically motorized intramedullary nail in congenital femoral deficiency. *J Pediatr Orthop*. 2019;39(9):458-465. doi:10.1097/BPO.0000000000001047
4. Bhave A, Baker E, Campbell M. Physical therapy during limb lengthening and deformity correction: principles and techniques. In: *Pediatric Lower Limb Deformities*. Springer; 2016:181-197.
5. Paley D, Guardo F. Lengthening reconstruction surgery for congenital femoral deficiency: fixator removal and rodding of femur. In: *Limb Lengthening and Reconstruction Surgery*. Springer; 2014:393-428.
6. Juliana BS, Fabio RB. *Fisioterapia Aquática Funcional*. Medical Arts; 2011.
7. Meysam R, Hamzeh B, Hamidreza A, Maria AFP. Effects of aquatic intervention on gross motor skills in children with cerebral palsy: a systematic review. *Phys Occup Ther Pediatr*. 2017;37(5):496-515. doi:10.1080/01942638.2016.1247938
8. Caromano APT, Themudo FMR, Canderolo JM. Physiological effects of immersion and exercise in water. *Fisioterapia Brasil*. 2005;4(1):60-65.
9. Edgar CP, Juliana MC. Benefits of hydrokinesiotherapy in patients with use of external fixator. *Biblioteca Uniban*. Published 2005. Accessed September 24, 2019. www.poolterapia.com.br
10. Wood GV. General principles of fracture treatment. In: *Campbell's Operative Orthopaedics*. 10th ed. Mosby CV; 2003:2560-2615.
11. Barllet DJ, Palisano R. Physical therapists' perceptions of factors influencing the acquisition of motor abilities of children with cerebral palsy: implications for clinical reasoning. *Phys Ther*. 2008;82(3):237-248.
12. Aidar FJ, Carneiro A, Silva A, Reis V, Garrido N, Vieira R. Cerebral palsy and aquatic activities: aspects linked to health and social function. *Motricidade*. 2006;2(2):109-116.
13. Norkin CC, Write DJ. *Medida do movimento articular: Manual de Goniometria*. 2nd ed. Medical Arts; 1997.
14. Pereira AC, Ribeiro MG, Araújo AP. Timed motor function tests capacity in healthy children. *Arch Dis Child*. 2016;101(2):147-151.
15. Morales NMO. *Cross-sectional Assessment of Quality of life in Children and Adolescents With Cerebral Palsy Using a Generic Instrument (CHQ-PF50)* [master's dissertation]. Universidade Federal de Uberlândia; 2005.
16. Simard S, Marchant M, Mencio G. The Ilizarov procedure: limb lengthening and its implications. *Phys Ther*. 1992;72(1):25-34.
17. Carregaro RL, Toledo AM. Physiological effects and scientific evidence of the effectiveness of aquatic physical therapy. *Rev Mov*. 2008;1(1):23-27.
18. Becker EB. Aquatic therapy: scientific foundations and clinical rehabilitation applications. *PM R*. 2009;1(9):859-872.

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