

Research article

Rehabilitation in Juvenile Idiopathic Arthritis with Hip Ankylosis

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Abstract: Introduction: Juvenile idiopathic arthritis (JIA) is the most common type of arthritis in children and teens. The inflammation of various joints causes pain. Early diagnosis and management of the patients, which include medication and rehabilitation therapies, can reduce the onset of complications. Despite the progress in managing this disease, there are cases which present severe complications. Material and Methods: A 13-year-old girl, diagnosed with JIA in a paediatric service in 2019, who is under treatment with Sulfasalazine since diagnosis, presented in the Paediatric Rehabilitation Centre of Băile 1 Mai with inflammatory pain in both knees, gait impairment with need of wheelchair, limited function in hands, elbows, shoulders, knees and hips which worsened in 2022, due to low compliance to the treatment and no check-up with a paediatric rheumatologist. She had 8 admissions in the Paediatric Rehabilitation Centre between October 2022 and July 2023. The examination reveals that there is a severe limitation of mobility in both hips and knees, the hands' small joints, elbows and shoulders. Laboratory tests were conducted showing no inflammatory markers. The radiographic finds sustained the changes encountered at the clinical examination. During her admissions in the Paediatric Rehabilitation Centre, she underwent a complex rehabilitation program which consisted of kinetotherapy, hydrokinetotherapy, massage, occupational therapy and physical agents. The program was designed according to the needs and the disease activity. Pharmacological means were used for the management of pain. The results were limited and the ability of standing up and walking were not recovered. Conclusion: In conclusion, JIA is a complex disease, which needs an early diagnosis and an early management program, which comprises of pharmacological treatment, rehabilitation program. Teamwork is necessary for managing JIA cases. The lack of compliance, and inadequate medication can lead to disabling complications.

Keywords: juvenile idiopathic arthritis; paediatric rehabilitation; hip ankylosis; polyarticular subtype.

1. Introduction

Juvenile idiopathic arthritis (JIA) is a chronic inflammatory disease, which affects the joints of children under 16 years old, lasts over 6 weeks and has unspecified aetiology. It is one of the most common chronic rheumatic diseases in children [1,2,3]. Patients with JIA usually present joint pain, accompanied by stiffness (morning stiffness especially), swelling and warmth, and avoidance of using the affected joint (this is more common in

children than complaints of pain), but they may have extra-articular manifestation also [2,3]. According to ILAR (International League of Associations for Rheumatology) there are six subtypes of JIA: Systemic JIA (sJIA), Polyarticular JIA (pJIA) with 2 forms: RF (Rheumatoid Factor) positive and RF negative, Oligoarticular JIA (oJIA), Enthesitis-related Arthritis (ERA), Psoriatic Arthritis (PsA) and Undefined Arthritis (UA) [1,3].

The diagnosis of JIA is a clinical one, but laboratory tests (blood cell counts, liver tests, kidney tests, ESR, CRP, RF, antinuclear antibodies) and imaging are used to exclude other pathologies, to fit the patient in the subtype and to assess the grade of activity and the extent of damage [3]. JIA is a highly debilitating disease. It is essential to have an early diagnosis in order to start the treatment and reach an inactive state as soon as possible. These will lead to an overall better outcome regarding the quality of life and the damage extent [4-6].

The main objectives of JIA treatment are: the management of pain, disease inactivity (or low active disease, if inactivity is not possible), reducing the joint damage, maintaining/restoring the normal function and activity level, normal growth and development, improvement of quality of life [1,2,4]. Treatment is individualised according to the JIA subtype, the activity level and personal needs of the patient. The pharmacological treatment consists of NSAIDs (nonsteroidal anti-inflammatory drugs), DMARDs (disease modifying antirheumatic drugs), glucocorticoids (oral and intra-articular) and biologic therapy [6]. Physical and occupational therapy, electro-physical agents, splinting, surgery (synovectomy, arthroplasty), balanced diet and psychological counselling are part of the non-pharmacological treatment [4,7].

2. Case report

A 13-year-old girl living in the countryside, from the Roma culture, without major physiological and pathological history, was admitted into our service for inflammatory pain in both knees, gait impairment, limited function in hands, elbows, shoulders, knees and hips. She has a sister diagnosed with autism. She is the second child in the family, from a physiological pregnancy, born at 38 weeks, spontaneously, with good adaptation to extrauterine life, vaccinated according to the national scheme. The menarche was at 12 years old. In 2016 the patient started to present gait impairment (out-toeing) and swelling of the joints of the hand and elbows. The disease progressed slowly over the years. In 2019, she started to accuse pain in the knees, with inflammatory characteristics and morning stiffness. Same year, she was diagnosed with Juvenile Idiopathic Arthritis in a paediatric service and treatment with Sulfasalazin was initiated and continues today (Sulfasalazin 500 mg 1-1-1 tb/day, oral). In 2021 she presented stature hypotrophy and the genetic karyotype was tested and no structural abnormality was found. In September 2002 she presented for rheumatology check-up and they didn't change the treatment, despite the progression of disease. She was guided to a paediatric rheumatology doctor, but she didn't go. In 2022, the gait impairment worsened suddenly, making the walking impossible. This conducted to the need of using a wheelchair. Since then, the patient has not been at school. In October 2022, she was admitted for the first time in the Paediatric Rehabilitation Centre of the Băile Felix Rehabilitation Hospital. She was admitted in our hospital for 8 times since October 2022 until July 2023.

After a thorough examination we discovered: 150 cm height, 37 kg weight, standing is possible with widened base of support, the knees bent and the flexion of the body on the thigh (Figure 1), walking is impossible, wheelchair is needed for mobility, Bouchard and Heberden nodes in both hands, metacarpophalangeal joint (MCP) swelling in both hands (Figure 2), positive Gaenslen sign in hands bilaterally, limited range of motion (ROM) in the small joints of the hand and in the wrists, finger opposition possible only with 2nd finger, 3rd and 4th finger with difficulty and 5th finger not possible on both hands, swelling of the both elbows, limited ROM of both elbows and shoulders, discreet

dorsal dextroscoliosis, discreet dorsal kyphosis, limited ROM in the cervical spine (limited extension, limited left rotation, limited lateral flexions, accentuated on the left side), both hips blocked in flexion at around 70° in left hip and 54° in right hip (Figure 3), limited ROM in both hips and knees, painful palpation of both knees, more accentuated on the left side, without pain at mobilisation of the knees, first metatarsophalangeal joint (MTP) swollen on both feet, hallux valgus on both feet, hammer toe on both 3rd toes, length of left leg 83 cm, length of right leg 82 cm, hypotrophy in the muscles of thighs and calves, patient's pain assessment using the visual analog scale (VAS) is 6. In Table 1 we present the angles assessed using goniometry. The other joints were within normal limits, and so were the other systems.



Figure 1. The patient in standing position with knees bent and the core bent over the thighs



Figure 2. Patient's hands with Bouchard and Heberden nodes and swollen metacarpophalangeal joints

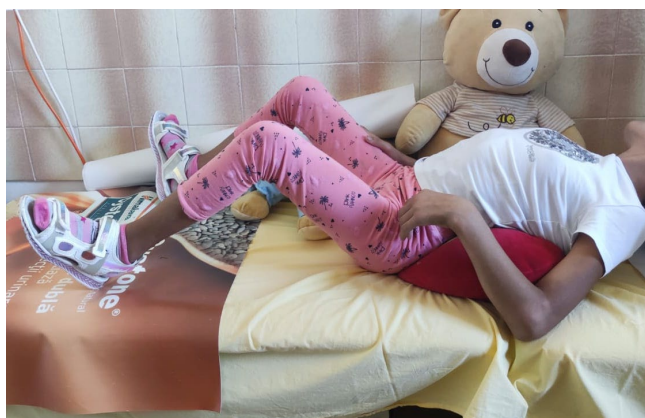


Figure 3. The patient's position is usually with bent knees and hips**Table 1.** Assessment of the active range of motion (ROM).

Joint and motion assessed	ROM ¹	Joint and motion assessed	ROM ¹
Left Shoulder		Left Hip	
Abduction	80°	Extension	10°
Flexion	64°	Flexion	Not possible
Right Shoulder		Abduction	Not possible
Abduction	85°	Right Hip	
Flexion	140°	Extension	8°
Left Elbow		Flexion	2°
Extension	120°	Abduction	Not possible
Flexion	70°	Left Knee	
Right Elbow		Flexion	130°
Extension	140°	Extension	40°
Flexion	80°	Right Knee	
Left Wrist		Flexion	142°
Flexion	55°	Extension	35°
Extension	25°		
Ulnar deviation	30°		
Radial deviation	<10°		
Right Wrist			
Flexion	38°		
Extension	20°		
Ulnar deviation	25°		
Radial deviation	<10°		

¹ROM= range of motion

We requested laboratory tests, which included the blood cell count, liver tests, kidney tests, inflammatory markers (ESR, CRP), Antistreptolysin O (ASO) titer, alkaline phosphatase and rheumatoid factor and the outcomes were usually within normal range. The ASO titer was increased (800 U/mL in October 2022 and 400 U/mL in November 2022). The ESR was normal (3 mm/h in October 2022 and 10 mm/h in November 2022), and so was the CRP (0,44 mg/dL in October 2022). The rheumatoid factor came out negative on two different tests. The alkaline phosphatase was increased in October 2022 (330.18 U/L).

We have also requested radiographies of the cervical spine, hands bilaterally, hips bilaterally and knees bilaterally. The positioning of the patient was difficult, because of the ankylosis of the hips. The cervical spine radiography (Figure 4) showed an increase of the cervical lordosis and reduced height of vertebral bodies C3-C6. The radiography of hands (Figure 5) showed diffuse osteoporosis in the carpal bones, carpo-metacarpal, metacarpo-phalangeal, proximal and distal interphalangeal on both hands, narrowing of carpal, the metacarpo-phalangeal and proximal and distal interphalangeal joints on both hands, distal epiphysis of proximal phalanges and medial phalanges have tumefied aspect, swelling of the soft tissue around proximal interphalangeal joints on both hands. bilateral hip radiography (Figure 6) presented narrowing of the joint space bilaterally, the joint space disappearing in the lower part of the joint, uneven articular surfaces on the right side, with osteocondensation, presence of marginal erosions and subcondral cysts, the femoral head is bilaterally deformed, flattened, with heterogenous aspect, more on the right side, no growth plates are visualised, aspect which suggests the end of ossification, pseudo-enlarging of the sacro-iliac joint space. The knee radiography (Figure 6) revealed narrowed joint space bilaterally with discret osteocondensation, more the tibial component of the joint.

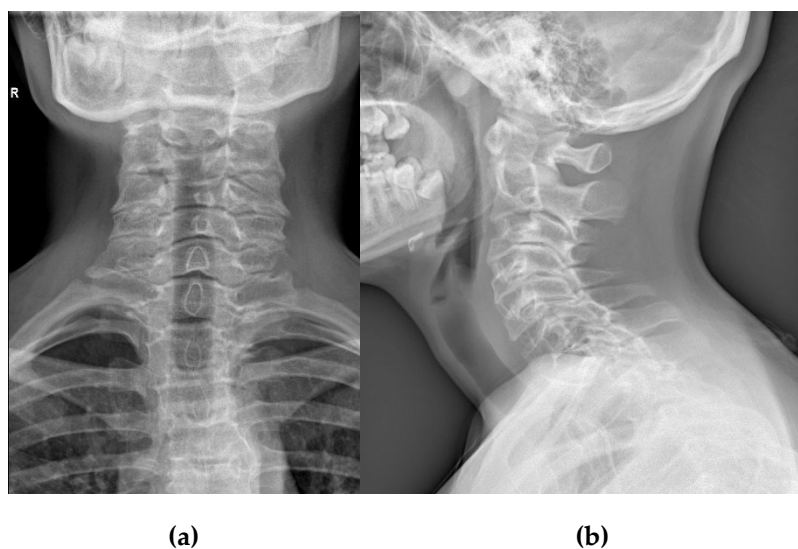


Figure 4. Cervical spine radiography- anteroposterior (a) and lateral view (b).

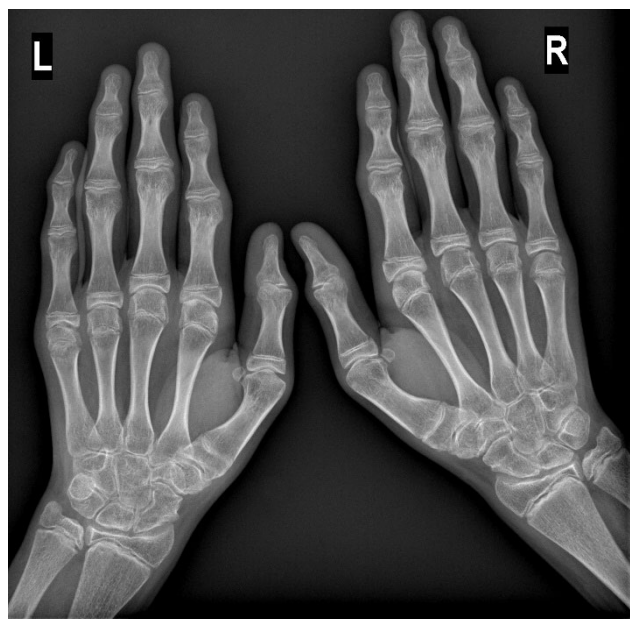


Figure 5. Bilateral hand radiography



Figure 6. Bilateral hip radiography



Figure 7. Radiography of the knees – antero-posterior view of right knee (a) and left knee (b) and lateral view of the right knee (c) and the left knee (d)

Using the Clinical Juvenile Activity Disease Activity Score 27 (cJADAS-27) we assessed the disease activity. The patient's assessment well-being measured on a VAS is 6 and the physician's assessment of patient's well-being on a VAS is 6. The final score was 26 out of a total of 47 or 4,57, meaning that the level of disease activity is moderate [8]. In this case was important to assess the damage the JIA produced. We used the Juvenile Arthritis Damage Index (JADI). It consists of two parts, measuring the joints (JADI-A) and extra-articular (JADI-E) damage. The JADI-A score is 15 out of a total of 72. The JADI-E score wasn't measured, because we didn't have an ophthalmology examination [9]. The functional status was assessed using the Childhood Health Assessment Questionnaire (C-HAQ). The score came out 1,03, which means that the patient's level of disability is mild-to-moderate [10].

According to the diagnostic and the assessment of the disease activity, damage and functional status, we have developed a treatment plan, which included both pharmacological and non-pharmacological means. The objectives were to reduce pain, increase joints' range of motion, increase the muscular strength, reduce the disability and improve the quality of life. The pharmacological treatment included the Sulfasalazine treatment

initiated in a paediatric service and non-steroidal anti-inflammatory drugs (NSAIDs), applied locally on the painful area and oral NSAIDs, when the pain level was high. The non-pharmacological treatment consisted of rehabilitation therapy, dietary counselling and psychological counselling. During the 8 admissions in the Paediatric Rehabilitation Centre, the rehabilitation program had small changes, according to the patient's needs at the moment of admission.

The main procedures that were part of the rehabilitation program were kineto-therapy, hydrokinetotherapy, massage and occupational therapy. The other procedures, which were changed to meet the patient's needs, were: lasertherapy, TENS, thermotherapy with paraffin, magnetotherapy, deep oscillation and electrical muscle stimulation for the quadriceps muscle and the triceps surae muscle. Mobilisation of the joints (both passive and active where was possible), muscle toning for the gluteus muscles, muscles of thigh and calves, and also for the muscles of the arm and forearm and muscle stretching. The objective of kinetotherapy was to increase or to maintain the ROM, and to increase the muscle strength in both legs and arms, so the patient would be able to transfer herself from the wheelchair to bed or where needed, and to increase the stability of the joints when the patient stands up. The stretching was made to reduce the muscle contracture and to increase the ROM joint. Hydrokinetotherapy was prescribed for the same reasons as kinetotherapy, and also for the ease of movement in the water due to the buoyancy and the myorelaxant effect of the thermal water in the Băile 1 Mai [11,12]. The lower limbs and the back were massaged for the myorelaxant and toning effect [13]. For maintaining or improving the hands' function, which mean gripping, prehension and finger opposition, occupational and recreational therapy was an important part of the rehabilitation program. Lasertherapy was applied on the right hip alternating with the knees in the first two admissions and later on was applied on the hands, for the anti-inflammatory and analgesic effect [14]. TENS was prescribed on the right hip alternating with the knees or the thoracic and lumbar paravertebral region for the analgesic effect. Because of the muscular hypotrophy of the thighs and calves, we applied electrical muscle stimulation for the quadriceps muscle and the triceps surae muscle [15]. Paraffin packs were applied on the hips, to help reduce the contracture of the pelvic belt muscles [11]. Magnetic field is used for its myorelaxant and sedative effect which is crucial for the main objectives of the rehabilitation program [15]. Deep oscillation is a therapy which combines deep vibration with massage and it was applied again for the myorelaxant effect to reduce the muscle contracture on the lower limbs and the hands [16].

Despite the numerous admissions, the results were limited. A slight improvement in the extension of the knees was seen (a few degrees). The muscle strength was also increased. An important outcome was the growth development. We have managed to maintain the ROM in the other joints. The hips ankylosis remained.

3. Discussion

JIA is a common chronic inflammatory disease amongst children, but the diagnosis is quite difficult because of the heterogenous clinical presentation. The advance of treatment in last decades made a huge impact on the evolution of the disease. But it is crucial to have an early diagnosis so the treatment can be started early and also a good compliance to the treatment [4,6]. Despite all these, there still are cases which get to the late stages of the disease and produce disability.

There are more factors which can lead to this outcome. For example, Drobnakova et al. [17] conducted a study amongst children in Slovakia and discovered that children of Roma culture have a higher risk of developing JIA and also more severe cases. Other factors that have been described to increase the risk of developing severe JIA are positive RF, positive anti-nuclear antibodies (ANA), hip involvement and symmetrical disease [1,3]. According to the data we collected from the patient, the subtype of JIA she has is

polyarticular with negative RF. We have no knowledge if she had the ANA tested. In our service, it was not possible to assess them. Because of the fact that she has a symmetrical disease pattern and both hips involved, the risk of a severe JIA is greater.

The severity of the disease in this patient, according to JADAS-27 and JADI is not that high at this point, but still, it created disability and produced ankylosis of the hips and knee flexum. In a previous check-up with a rheumatologist, it was stated that a treatment change is not necessary as long as the laboratory tests didn't show inflammation. Subclinical disease activity may be present in patients with normal inflammatory markers [18], and this may lead to progression of the disease. This may be one of the reasons why the patient's disease progressed, despite the treatment with Sulfasalazine. Another reason for disease progression might be low compliance to the treatment and the fact that she didn't present for the follow-up. The patient stated that before the walking became impossible, she might have skipped the midday dose in some days. The patient presented intermittent pain in the knees and hips which determined the patient to adopt positions to reduce the pain [4, 19, 20]. Together with the inflammatory process and the sparing of the joints, it might have led to muscle contractures and the changes in the joints that we saw in the radiographs.

Another frequent consequence of JIA is the growth and puberty delay, due to the inflammation, treatment with corticosteroids, physical inactivity, muscular mass decrease, undernutrition caused by pain [21]. Our patient has normal height (151 cm) and weight (38 kg) for her age, according to the growing charts [22]. We must mention, that in May 2022, patient was admitted in the ER and the weight was 16 kg, which was under the normal values for her age. An improvement in the patient's weight was seen during 8 admissions in the Paediatric Rehabilitation Centre, presenting a general growth of about 8 kilograms. Another important thing is the puberty. The patient's menarche was at 12 years old, which is within normal age range for girls, according to the Tanner stages [23]. Asymmetrical growth development is another consequence of JIA, especially in the oligoarticular subtype. It means that there might be discrepancies in the limbs' length [21]. Our patient presented 1 cm difference between the lower limbs, left leg being longer than the right leg, which doesn't create a big impact.

The presentation in the Paediatric Rehabilitation Centre was a little late for the walking disability. But it is still crucial for the improvement of quality of life and in achieving independence (as much as possible) in the daily life activities [2, 11]. The American College of Rheumatology stated in the 2021 Guideline for JIA treatment, that physical therapy is recommended regardless the pharmacological approach [7]. The main goals of the rehabilitation programs in this case were to increase or maintain the ROM and to increase the muscle strength for joint stability and for transfers. The complex program which included kinetotherapy, massage, hydrokinetotherapy and physical agents, was designed according to the disease status, as it is recommended [2, 11]. Most studies showed that a rehabilitation program is necessary in avoiding the installation or progression of disability and it also increased the quality of life of the patients. Better outcomes were seen in patients who underwent programs similar to the program we indicated highlighting the importance of kinetotherapy and hydrokinetotherapy, rather than only one or two procedures which didn't include exercises. Another problem amongst children with JIA is the low physical condition compared to her healthy peers. That's why an active life which comprises daily exercises is recommended [24-27]. It's important for the patients to continue at home the exercises they learn in rehabilitation centres, for a better outcome. In our case it is highly improbable that the patient doesn't exercise at home. Serial casting might be a solution to the limited ROM in the knees of our patient [11, 25].

The involvement of the hip is not necessarily common in JIA, but it is more frequent in adults with rheumatoid arthritis [19]. It is a risk factor for developing more severe disease [3]. Aguiar et al. [28] presented the case of a girl with structural damage in the hip

which regressed under biological treatment. Calafi et al. [29] described a case of bilateral destructive hip disease in a girl who was not following any treatment and which conducted to complete destruction of the femoral head. Our patient presents structural damage in both hips which determined the ankylosis of the joints. Teamwork is necessary for a future management of this case. A paediatric rheumatologist, a paediatric orthopedic surgeon and a physiatrist should be part of this team. A change in the pharmacological treatment might determine a good outcome in this case, just like the case presented by Aguiar et al. [28]. Another possibility is undergoing a total hip arthroplasty (THA) in both hips. Van de Velde et al. [30] found that THA before the age of 16 is correlated with lower pain levels and improvement of the ROM and walking. The timing is important when THA is considered, because children still have growth plates, but the impact of the pain is also a decision factor [4]. Perioperative management is also crucial. It includes pharmacological management (possible interruption of the treatment), the selection of the implant (custom-made implants might be required, due to the skeletal immaturity) and the rehabilitation program [4]. Our patient is at a young age, and a major surgery might be postponed. Until then, rehabilitation program is key to maintaining the strength of the muscles and the other joints' ROM and stability.

School dropout is more frequent amongst children with JIA, than in their healthy peers, as Bouaddi et al. [31] showed in their study. They stated that the disease activity and functional impairment were one of the factors which determined school absenteeism. Our patient said that the disability which requires the use of the wheelchair is one of the main reasons why she didn't go to school.

4. Conclusion

JIA, despite the latest advances, still remains a challenging pathology. Delayed diagnosis and treatment can lead to progression of the disease. Also, the lack of compliance conduct to disability onset. An early rehabilitation program determines fewer joint deformities, functional limitations and muscle atrophies. Even though the patient presented in a late stage of the disease, the quality of life was improved after rehabilitation program. The complexity of it also increased the limited success of the rehabilitation program. For better outcomes, the patients should be sent earlier to the rheumatologist and physiatrist. The follow-up is crucial for treatment adjustment. Teamwork remains the key for better results in diseases like JIA.

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