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Rehabiliation vs Surgery for Recurrent Snapping Hip in A 26-Year-Old Man with Joint Hypermobility Syndrome/Ehlers-Danlos Syndrome Hypermobility Type: A Biomechanical Study

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Abstract

Recurrent snapping hip is a common clinical presentation of localized joint hypermobility and it is presumed to be more common in patients with generalized forms of joint hypermobility, such as joint hypermobility syndrome/Ehlers-Danlos syndrome, hypermobility type (JHS/EDS-HT). This case study aimed was to show the effects of rehabilitation on a 26-year-old man with recurrent snapping hip and JHS/EDS-HT. He previously underwent surgery only to experience complete recurrence after one year. The patient followed a four month treatment program consisting of passive and assisted-active mobilization, joint stabilizing and proprioceptive enhancement exercises, and he was examined and evaluated before and after the program using clinical scales and a three-dimensional (3D) motion analysis system. The treatment led to improved scores on all clinical scales and pain assessment measures. Motion analysis showed more physiological spatio-temporal parameters and biomechanical pattern at the proximal joints during walking following treatment, while the ankle joints appeared more impaired in terms of the physiologic pattern. Our observations demonstrate the beneficial effects of rehabilitation in the mid-term management of snapping hip when it recurs after surgery in JHS/EDS-HT. These findings support the general concept of low efficacy of orthopedic surgery in JHS/EDS-HT and furthermore they highlight the benefits of physical therapy in the management of this syndrome in JHS/EDS-HT patients.

Keywords: Hypermobility; Ehlers-Danlos syndrome; Gait analysis; Physical disabilities, Rehabilitation; Snapping hip

Introduction

Ehlers-Danlos syndrome (EDS) is an umbrella term for a clinically variable and genetically heterogeneous group of inherited connective tissue disorders that are primarily characterized by the following three symptoms: joint hypermobility, skin hyperextensibility and tissue fragility [1]. On the basis of available diagnostic criteria [2], six major types of EDS and some additional rarer variants have been identified [3]. The hypermobility and the classic types are the most common, accounting for approximately 90% of all EDS cases [4]. Awareness of these disorders and, more specifically, the hypermobility type of EDS (EDS-HT), is growing due to their recently outlined disability potential [5]. Though it is still considered a rare disorder, the perceived rate of EDS-HT is increasing also because it is currently considered the same as joint hypermobility syndrome (JHS) [6]. Recognizing JHS/EDS-HT is a difficult task since it is one of the few EDS that does not have known molecular basis; so therefore it is an exclusion diagnosis [7]. Consequently, misdiagnoses and lack of diagnoses are serious problems for these patients [8].

JHS/EDS-HT is mainly characterized by joint instability that may be associated with a wide variety of potential consequences. In JHS/EDS-HT, congenital joint laxity leads to joint instability and a predisposition to ligamentous injuries, joint effusions, subluxations and dislocations. This devastating progression often leads patients to consider orthopedic surgery [9]. Although, in a cohort of 30 EDS patients, the estimated rate of surgeries per patient and sites for surgery per patient were 1.17 and 2.06 respectively [9]. However very little is known about the mid- and long-term follow up of orthopedic surgery in these patients. Recurring symptoms and side effects from surgery are probably higher in JHS/EDS-HT, as demonstrated by a questionnaire study on patients with different types of EDS (many with JHS/EDS-HT) that shows musculoskeletal pain associated with previous surgery [10].

In this study, we discuss a 26-year-old man with JHS/EDS-HT and snapping hip. He was unsuccessfully treated with surgery for his right snapping hip at 18 years of age while similar features appeared contralaterally some years later. We critically analyzed possible therapeutic options and outlined a personalized rehabilitation program that led to promising results, as demonstrated by using clinical scales and 3D quantitative analysis of walking, or 3D Gait Analysis (3D-GA) to assess its effects.

Patient and Methods

Case description

A 26-year-old male patient with EDS was referred to our rehabilitation center. The patient provided written informed consent before participating in the study. He reported severe pain in his hips while at rest and during active mobilization associated with walking impairment that causes him to walk with two crutches to avoid the load.

His past medical history revealed generalized joint instability associated with other symptoms including spontaneous skin bruising, gingival fragility and recurrent dental caries, chronic asthenia and fatigue, abdominal pain, myalgias and poly articular recurrent pain since he was a young child. There did not appear to be other similar cases in his family. Psychomotor development was normal and family history was negative.

When he was 18 years old, , he began to experience major pain associated with an audible snapping in his right hip when getting up from a seated position on the sofa; he subsequently began to experience such pain and hip snapping about once a month. An X-Ray examination and an MRI of the right hip were performed and consequently a diagnosis of "snapping hip" or "coxa saltans" was made; the patient's blood was negative for inflammatory markers or autoantibodies. There was not sufficient evidence of inflammatory joint disease and the electrophysiological examination was negative.

Because of the persistence of the pain and the recurring snapping, the patient underwent "z-plasty of iliotibial band" surgery. This technique is used to lengthen the tight iliotibial tract and to shift the thickened band anteriorly so that it no longer flicks over the great trochanter as the hip is flexed. After surgery, the patient was able to walk for one year without pain only to then experience another dislocation while getting out of a car and this led him to seek out revision surgery.

The absence of pain, improved walking and engaging in physical activities like swimming lasted less than one year when the patient's back and hip pain reappeared; these symptoms were followed, in a short time, by a new episode of snapping and pain of his left hip that made him walk with two crutches.

At physical examination, limitation in passive movement of the hip associated with pain was present bilaterally greatest on the left. The hips range of motion were 80°for the passive flexion, 20° for the abduction , the intrarotation was impossible for pain and the extra-rotation was 20°. On the right side the passive flexion was 100°, abduction was 40°, intra-rotation was20° and extra-rotation 30°. The patient had soft and velvety skin a keloid scar on the hip area related to the surgical cut. The Beighton score was 4/9 (bilaterally elbow hyperextension and bilaterally thumb opposition); the bending test and the hyperextension of knees were not necessary for the pain presented by the patient. The neurological examination did not reveal any sensory or motor abnormalities. In the absence of any overt sign of other hereditary connective tissue disorders, the diagnosis of JHS/EDS-HT was established in accordance with the Villefranche (EDS-HT) (2) and Brighton (JHS) [3] criteria.

Assuming that joint instability is secondary to joint hypermobility and to tendon hyper-flexibility, we outlined a rehabilitation program to improve his muscle strength, to recover the range of motion for his hip, and to reduce pain. The rehabilitation program consisted of two 60 minute sessions performed on separate days each week: one involved walking and the other involved a physical activity in a swimming pool. This treatment consisted of: (i) passive and assistive-active mobilization of the joint, only in the physiological range of articular motion; (ii) joint stabilizing exercises to reduce "excessive" joint range; (iii) exercises for proprioceptive enhancement; and (iv) passive and active mobilization of the hip, pelvis and lumbar spine in accordance with the Maitland technique.

Recording measures

The patient was evaluated by clinical scales and 3D-GA before (PRE) and at the end (POST) of the proposed four-month rehabilitation program. The following scales were used:

- Lower Extremity Functional Scale (LEFS), for the assessment of lower extremity musculoskeletal dysfunction [11];
- the Harris Hip Score (HHS), for hip features [12];
- the Tinetti Balance (TB) and Gait (TG) Evaluation for walking and balance [13]; and
- the Numeric Rating Scale (NRS) to estimate pain intensity [14].

His gait pattern was quantitatively evaluated using a 12-camera optoelectronic system (ELITE 2002, BTS, Milan, Italy) with a sampling rate of 100 Hz, two force platforms (Kistler, CH) and a 2 TV camera Video system (BTS, Italy) that was synchronized with the system and the platforms for video-recording (BTS, Italy); after placement of passive markers on the patient's participant's body, as described by Davis [15], the patient was asked to walk barefoot at his own natural pace (self-selected speed) along a walkway containing the force platforms at the mid-point. At least five trials were collected for each session in order to ensure the consistency of the data. All graphs obtained from the 3D-GA were normalized as a percentage of gait cycle; after analyzing the graphs, we identified some parameters (time/distance parameters, joint angle values at specific instances of the gait cycle). It was not possible to gather kinetic data since the patient was not able to properly place one foot on each platform, especially in the PRE session. Although various parameters were calculated, we chose to further examine the most reliable and consistent ones for analyses in individuals with EDS [16,17] and they are listed below as follows:

Spatio-temporal parameters

a) % stance: duration of the stance phase (as % of the gait cycle);

b) Step length: longitudinal distance from one foot strike to the next one (mm);

c) Step width (mm); and

d) Velocity: mean velocity of progression (m/s).

Kinematics

a) The mean value of pelvis on the sagittal plane (Mean PT index), expressed in degrees;

b) The values of angle of ankle (AIC index), knee (KIC index) and hip joint (HIC index) upon contact of the foot with the ground (i.e. Initial Contact or IC), expressed in degrees;

c) The values of maximal ankle dorsiflexion during stance phase (AMSt index) and the maximal flexion of the knee (KMSw index) during swing phase, expressed in degrees;

d) The values of minimal ankle dorsiflexion at toe-off (AmSt index), knee (KmSt index) and hip flexion (HmSt index) during the gait cycle, expressed in degrees; and

e) The range of motion of the pelvis on the coronal (PO-ROM index) and transversal (PR-ROM index) plane; the range of motion of hip on coronal (HAA-ROM index) and sagittal (HFE-ROM) plane; the range of motion of knee (KFE-ROM index) on sagittal plane; the range of motion of ankle on sagittal plane during stance phase (ADP-ROM index), expressed in degrees.

All the previously defined parameters of 3D-GA were computed for all trials and then the mean values and standard deviations of all indexes were calculated for the patient's right and left side for each session. All the computed parameters were compared with the values of an age-matched control group (CG) [16].

After evaluating the fit of the observed data to normal distribution (Kolmogorov-Smirnov test) and homogeneity of variances (Levene's test), the t-test for dependent samples and the t-test for independent samples were applied to compare PRE and POST sessions and to compare each session with the CG. Null hypotheses were rejected when probabilities were below 0.05.

Results

After four months of rehabilitation, the progressively was once again able to walk without any devices and he experienced a major of pain intensity. All the functional scores (LEFS, HHS, TB and TG) increased their values and the pain score (NRS) decreased, indicating a great improvement after the rehabilitation program, as shown in Table 1.

	PRE	POST	% Variation
LEFS	17	70	+312%
HHS	10	9	+840%
TB	4	14	+250%
TG	3	12	+300%
NRS	9	1	-89%

Table 1: Functional scale scores (LEFS, HHS, TB, TG) and NRS score for pain before (PRE) and after (POST) the rehabilitation program and percentage of variation (%variation).

LEFS: Lower Extremity Functional Scale; HHS: Harris Hip Score; TB: Tinetti Balance; TG: Tinetti Gait; NRS: Numeric Rating Scale.

As far as 3D-GA, in Table 2 the mean (standard deviation) values for each index computed in the two sessions (PRE and POST) and for each limb are listed. As far as spatio-temporal parameters, before treatment (PRE session) the patient walked with a slower progression, longer bilateral stance time, reduced step length, particularly on the right side, and larger step width, when compared to normal values. In the POST session all these parameters improved as values were closer to the norm.

	PRE		POST		CG
Spatio-temporal parameters					
	Right	Left	Right	Left	
%stance (% gait cycle)	79.0 (5.7)+	73.5 (4.9)+	57.0 (2.8)*	56.1 (2.5)*	59.6 (3.2)
Step length (mm)	115.2 (4.2)+	434.5 (7.1)+	582.5 (16.2)*	588. (14.8)*	556.2 (42.4)
Step width (mm)	182.0 (4.2)+		141.1 (7.1)*		123.3 (27.8)
Velocity (m/s)	0.2 (0.1)+		0.9 (0.1)*		1.1 (0.2)
Pelvis (°)					
Mean PT	28.4 (1.2)+	29.2 (1.7)+	7.4 (0.6)*	8.5 (0.7)*	6.5 (6.9)
PO-ROM	7.6 (1.1)	8.2 (1.2)	4.9 (1.5)	4.4 (1.0)	6.0 (2.5)
PR-ROM	7.7 (1.9)	7.2 (1.1)	9.6 (0.3)	9.5 (0.1)	10.7 (5.3)
Hip joint (°)					
HIC	33.4 (4.1)+	40.1 (0.9)+	25.6 (3.6)*	24.1 (0.6)*	27.2 (9.6)
HmSt	5.3 (0.5)+	19.2 (4.6)+	-12.7 (2.2)*	-10.2 (4.4)*	-14.8 (9.6)
HFE-ROM	32.5 (3.5)+	27.5 (2.3)+	40.1 (0.7)*	39.5 (3.6)*	43.5 (4.8)
HAA-ROM	9.9 (3.1)	7.2 (1.4)	6.9 (0.9)	6.2 (1.8)	10.7 (3.1)
Knee joint (°)					
KIC	5.6 (3.3)	-1.2 (0.5)	4.4 (2.3)	-1.0 (0.1)	4.4 (5.5)
KmSt	-0.6 (1.2)	-2.6 (1.0)	-2.8 (2.1)	-1.2 (0.2)	0.1 (3.8)
KMSw	41.7 (3.5)+	26.4 (4.5)+	55.1 (3.8)*	57.0 (2.8)*	57.1 (6.4)
KFE-ROM	42.3 (7.4)+	28.3 (4.7)+	57.8 (5.9)*	59.0 (6.1)*	59.1 (6.3)

Ankle joint (°)					
AIC	0.2 (0.1)	-8.7 (1.8)+	-4.9 (0.7)*+	-10.9 (3.3)+	1.8 (4.9)
AMSt	11.8 (0.3)+	3.7 (1.7)+	-0.3 (1.3)*+	4.1 (1.8)+	21.0 (5.2)
AmSt	-4.4 (0.2)+	-8.0 (1.8)	-31.3 (0.3)*+	-26.3 (1.7)*+	-9.7 (9.4)
ADP-ROM	16.2 (0.1)+	11.8 (3.6)+	30.9 (1.0)*	30.4 (0.1)*	25.7 (6.6)

Table 2: Spatio-temporal and kinematic parameters of the evaluated patient in the PRE and POST session and for CG. *= p< 0.05, PRE vs. POST session; += p< 0.05, PRE/POST vs. CG.

Data are expressed as mean (standard deviation).

(ROM: Range of Motion; PT: Pelvic Tilt; PO: Pelvic Obliquity; HIC: Hip at IC; HFE: Hip Flex-Extension; HAA: Hip Ab-Adduction; KIC: Knee at IC; KFE: Knee Flex-Extension; AIC: Ankle at IC; ADP: Ankle Dorsi-Plantarflexion; IC: Initial Contact; St: Stance; Sw: Swing; M: maximum value; m: minimum value; CG: Control Group).

In terms of joints kinematics, the pelvic tilt, which was characterized by an anterior position on the sagittal plane (Mean PT index), reduced its ante version position, approaching a value close to CG. No changes occurred on the frontal and transversal Planes (PO-ROM and PR-ROM indices), which remained similar to normal values. Before treatment the hip joint appeared flexed during the whole gait cycle, especially on the left side; the patient walked with an excessive flexion at initial contact (HIC index) and reduced extension ability in mid-stance (HmSt index), causing a bilateral reduction of joint excursion (HFE-ROM index). In the POST session, the HIC and HmSt indices decreased at both limbs, with more physiological values closer to the CG. No changes occurred on the frontal plane (HAA-ROM index) which remained close to normal. The knee joint in PRE session was characterized by quite a normal position in the stance phase (KIC and KmSt indices), even if the left side was slightly more extended at initial contact; in the swing phase, a bilateral reduction of the knee flexion (KMSw index) was evident, leading to a limited joint excursion (KFE-ROM index), especially on the left side. The intervention led to bilateral improvement in the swing phase, highlighting an increase of knee flexion and a better range of motion similar to the CG. In terms of the ankle joint, the right side was characterized by quite a normal position at initial contact (AIC index), with a reduced range of motion (ADP-ROM index) due to the reduced dorsiflexion in the stance phase (AMSt index) and the limited plantar flexion ability at the toe-off (AmSt index) when compared respect to healthy subjects; the left side exhibited a plantar flexion position at initial contact (AIC index) and a reduced dorsiflexion in the stance phase (AMSt index), with a lower joint excursion than the CG. After the treatment, the patient showed some significant changes on the right side, which was more plantar flexed at initial contact and in stance phase, and bilaterally at the toe-off, which increased the plantar flexed position; consequently, the ranges of motion increased, with values closer to normal.

After one year, the patient was telephonically interviewed, because he was abroad, and he reported good health and the continuation of his improved condition.

Discussion

Snapping hip is an audible and sometimes painful disorder of the hip that occurs during activities that require repetitive flexion, extension and abduction of the hip [18]. Its causes can be classified as external, internal, and intra-articular. The most common type is the external one, in which the ileotibial band or the gluteus maximum slides over the greater trochanter during repetitive flexion and extension [19]. Asymptomatic snapping hip should be considered a benign and normal occurrence, especially in athletes [18]. However, it may sometimes cause pain and interfere with the patient's ability to perform maneuvers that stretch the iliotibial band and/or gluteus maximum tendon over the greater trochanter [20]. Treatment is usually conservative and involves stretching, physical therapy and/or anti-inflammatory drugs. Surgery is usually considered in refractory cases only. The z-plasty lengthening of the ileotibial band is recommend in literature as the primary surgical modality for relief in the refractory, yet symptomatic, snapping hip due to a tight iliotibial band [21]. Snapping hip, especially in its external variant, may be considered a common clinical presentation of localized joint instability. Early studies found a high rate of snapping hip among EDS patients [22] We believe that many of the unclassified patients may be affected by the common JHS/EDS-HT variant.

In relevant literature, rehabilitation treatment is often described as the mainstay of treatment for the musculoskeletal aspects of this pathology, but physiotherapists have frequently admitted that they did not know how to best help their patients [23]. The main difficulty is understanding the main problems of these patients that have chronic pain that is unsuccessfully treated using conventional methods; they also have diminished joint proprioception with an impaired position sense that leads to difficulties in movement that are often associated with pain.

Even if surgical results in snapping hip management are described as excellent and predictable [21], in EDS patients such interventions are often unsuccessful. This may be likely due to the connective properties.

In this case report we quantitatively demonstrated the typical gait pattern of "snapping hip" from 3D-GA plot, characterized by high hip flexion during the entire gait cycle with a reduced ROM in comparison with the CG; secondly we quantified the changes in terms of pain and functional scores and in terms of walking ability in a patient with EDS after a rehabilitative program. Our results underlined improvements on all the clinical scales, which mean a reduction of pain and functional improvement. As far as of gait ability we can observe improvements in terms of spatio-temporal parameters and kinematics, especially at the hip and knee joints, which increased their ranges of motion. On the contrary, as far as ankle kinematics, a slight change appeared especially at the right side which was in a more plantar flexed position during the entire gait cycle. The most important result the improvement at proximal level, and in particular at hip joint, that was the main goal of the rehabilitative program; the changes at ankle joint may be considered a strategy to for stability after the modifications at the proximal joint following the rehabilitation program which led to a new gait strategy.

The use of 3D motion analysis successfully quantified the functional limitation in a patient with EDS and the effects of the proposed a personalized rehabilitation program which seems to generate promising results. Using the 3D-GA it was possible to investigate in detail the motor abilities required in daily activity such as walking. The kinematic data adds a whole new set of information that can more accurately characterize the functional limitation of a patient with snapping hip and the changes after rehabilitation program, suggesting an intensification of rehabilitation program mainly at distal joint, as ankle.

The 3D-GA allows for continuous monitoring of a subject and it could be used during future applications to provide guidance for a more specifically focused physical therapy. The quantitative graphs and the numerical parameters collected using the 3D-GA can help healthcare providers to accurately target and improve the therapeutic interventions.

In brief, data observed with 3D-GA supports the general concept of favoring the option physical therapy in JHS/EDS-HT as a suitable and efficacy treatment not only when surgery may have failed but in all JHS/EDS-HT, as a primary form of first line treatment. Moreover the exact characterization of the gait pattern may help to better plan the rehabilitation program.

This study has some limitations. Only a single case is reported and the analysis was limited to only kinematic data. However, this is the first paper to use a 3D motion analysis system to quantitatively and objectively assess the functional limitation(s) experienced by a patient suffering from EDS with snapping hip. Further studies should be conducted on a wider group with the same disease and they should analyze gait pattern and monitor the evolution of motor skills over time.

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