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# Kinematic and plantar pressure analysis in Strumpell-Lorrain disease: A case report

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ARTICLE INFO	A B S T R A C T		
A R T I C L E I N F O Keywords: Gait abnormalities Spasticity Strumpell-Lorrain Hereditary spastic paraparesis Lower limb weakness	Background: Hereditary Spastic Paraparesis (HSP), also known as Strumpell-Lorrain disease, is a neurodegener- ative disorder characterized by progressive muscle weakness, lower limb spasticity, and abnormal gait. It is a genetically inherited condition affecting the spinal cord. Currently, there is a lack of scientific literature on the evaluation of space-time parameters and plantar pressures in individuals with HSP. Therefore, the objective of this study is to assess spatial-temporal parameters and plantar pressures using motion sensors and a bar- opodometric platform. <i>Case presentation:</i> A 50-year-old female patient with a body mass index (BMI) of 22.28 presented to our hospital with a 12-year history of Strumpell-Lorrain disease. She exhibited typical symptoms of the disease, including spastic paraparesis in the lower limbs, leading to difficulty in walking. The initial symptom of difficulty walking was diagnosed when she was 38 years old. Apart from walking impairments associated with spasticity and ataxia, the patient demonstrated paraparesis and weakness in the lower limbs, without any cognitive deficits. <i>Conclusions:</i> This analysis elucidates the challenges faced by the patient with Strumpell-Lorrain disease in walking, particularly during the swing phase, resulting in a reliance on monopodal support. Additionally, the patient experiences difficulty in dorsiflexion of the ankle due to spasticity in the gastrocnemius muscle, leading to an increased load on the forefoot. These findings contribute to a better understanding of the specific gait ab- normalities and biomechanical impairments associated with Strumpell-Lorrain disease.		

#### Introduction

Strumpell-Lorrain disease, also known as hereditary or familial spastic paraplegia (HSP or FSP), is a group of rare neurological disorders that primarily affect the upper motor neurons, resulting in stiffness and weakness in the legs [1,2]. FSP is estimated to affect 7.4 in 100,000 individuals and is inherited in a genetic manner, passed down through generations. The autosomal dominant (AD) form of FSP is the most common, accounting for 70–80% of all cases [1–3]. The main characteristics of hereditary spastic paraplegia include varying degrees of muscle stiffness, muscle spasms, weakening of leg muscles, and bladder control problems. Some families may also experience more severe symptoms such as mental retardation, dementia, epilepsy, peripheral neuropathy, retinopathy, deafness, ataxia, dysarthria, and disorders of the extrapyramidal system [4].

The onset of the disease is typically gradual, slow, and insidious, with symptoms worsening progressively over time. The age at which symptoms appear can vary greatly among different families and even within the same family [2–4]. Diagnosis of hereditary spastic paraplegia is typically based on a thorough evaluation of the patient's personal and family history, a comprehensive physical examination, and assessment of characteristic symptoms and findings. Specialized tests such as neurophysiological or genetic studies may also be conducted for diagnostic purposes [5].

The treatment of hereditary spastic paraplegia focuses on managing symptoms and includes medical interventions and physiotherapy. Currently, there is no treatment available that can slow down or modify the progression of the disease, although baclofen may help reduce spasticity in some patients [1,2,4].

In this particular pathology, the evaluation of space-time parameters and plantar pressures has not been previously conducted. These assessments, using motion sensors and a baropodometric platform, can provide valuable information for rehabilitation and habilitation in degenerative diseases. The aim of this study is to assess spatial-temporal

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Fig. 1. Static baropodometry platform: plantar pressures in orthostatism.

parameters and plantar pressures using these techniques. In recent years, there has been growing interest in utilizing advanced technologies such as motion sensors and baropodometric platforms to evaluate spacetime parameters and plantar pressures in various degenerative diseases. However, the application of these assessments in the context of Strumpell-Lorrain disease, also known as hereditary spastic paraplegia (HSP or FSP), has not been extensively explored [1].

Therefore, the aim of this study is to investigate the potential benefits of utilizing motion sensors and a baropodometric platform to assess spatial-temporal parameters and plantar pressures in individuals with hereditary spastic paraplegia. By utilizing these techniques, researchers and healthcare professionals hope to gain valuable insights into the gait characteristics, foot mechanics, and pressure distribution patterns of individuals affected by this rare neurological disorder [2].

The evaluation of space-time parameters, including stride length, cadence, and gait velocity, can provide essential information about the walking pattern and mobility of individuals with hereditary spastic paraplegia. Understanding these parameters can aid in the development of targeted rehabilitation and habilitation strategies, with the goal of improving walking ability and overall functional independence.

Additionally, the assessment of plantar pressures can offer valuable insights into the distribution of forces exerted on the feet during walking. This information can help identify areas of excessive pressure or abnormal loading, which may contribute to the development of foot deformities or ulcers. By identifying these pressure distribution patterns, healthcare professionals can develop customized interventions, such as orthotic devices or specific footwear, to alleviate discomfort, prevent complications, and improve overall foot health.

It is worth noting that this study represents a novel approach in the evaluation and management of hereditary spastic paraplegia [3]. If successful, the findings from this research could potentially contribute to the development of more tailored rehabilitation and habilitation programs for individuals with this rare neurological disorder. Furthermore, the utilization of motion sensors and a baropodometric platform may have broader implications in the assessment and treatment of other degenerative diseases affecting gait and foot function.

#### **Case presentations**

A 50-year-old female patient with a BMI of 22.28, who has been suffering from Strumpell-Lorrain disease for 12 years, presented at our renowned hospital seeking treatment. The patient exhibited typical symptoms of the disease, including difficulty walking and spastic paraparesis in the lower limbs. The onset of walking difficulties occurred at the age of 38, and since then, her mobility has progressively declined, characterized by spasticity, ataxia, paraparesis, and weakness in the lower limbs. However, there have been no indications of cognitive decline.

In order to gain a better understanding of the patient's condition and provide appropriate treatment, she provided informed consent and underwent evaluations utilizing a baropodometric footboard and Wiva Science inertial motion sensors. The baropodometric footboard was employed to assess the patient's plantar pressures under both static and dynamic conditions. The motion sensors were utilized to analyze spatiotemporal parameters, including speed, cadence, and timing during a prescribed walking protocol. This comprehensive evaluation has yielded valuable insights into the patient's condition, which will aid in the development of effective rehabilitation therapies aimed at improving her walking abilities.

#### **Clinical findings**

A 50-year-old female patient with a BMI of 22.28, diagnosed with Strumpell-Lorrain disease (SLD) 12 years ago, presented at our renowned medical facility seeking specialized treatment. The patient exhibited characteristic features of SLD, including spastic paraparesis and impaired locomotion due to progressive muscular weakness and lower limb stiffness. The onset of ambulatory difficulties occurred at 38 years of age, with subsequent gradual deterioration of mobility accompanied by spasticity, ataxia, and weakness in the lower extremities. Notably, no cognitive deficits were observed.

Following informed consent, a comprehensive assessment was performed utilizing a baropodometric footboard and Wiva Science inertial motion sensors to obtain a thorough understanding of the patient's



Fig. 2. Dynamic baropodometry platform: plantar presses during walking.

condition and facilitate individualized therapeutic interventions. The baropodometric footboard enabled the evaluation of plantar pressure distribution in both static and dynamic contexts, providing insights into pressure patterns during different phases of gait. The motion sensors precisely quantified spatiotemporal parameters, including gait velocity, cadence, and temporal sequencing, during a standardized walking protocol.

The integration of these advanced technologies yielded objective and precise data, enabling a comprehensive analysis of the patient's gait characteristics and foot biomechanics. This detailed assessment serves as a foundation for the development of targeted rehabilitation strategies aimed at improving ambulatory function and overall functional independence.

The comprehensive evaluation employing state-of-the-art technologies represents a significant advancement in the management and care of individuals affected by SLD. The obtained findings will inform the design of effective and personalized rehabilitation interventions, with the ultimate goal of enhancing mobility and quality of life for the

#### Table 1

Results table with wiva sensors: view sensor results with time up and go protocol.

Parameter	Patient (right side)	Patient (left side)	Healthy individual (average)
Maximum Pressure	303.8 KPa	292.1 KPa	250–400 KPa
Velocity	1480 ms	1320 ms	1200–1500 ms [10,11]
Cadence	-	50.5	55–115
Gait Cycle	-	11 s	0.8–1.2 s
Double Support Time	-	15.7 ms	10–30% of gait cycle
Single Support Time	-	7 ms	20-40% of gait cycle

patient.

#### **Diagnostic assessment**

#### Baropodometric footplate

The baropodometric platform is an instrument used in a clinical setting to quantify plantar pressure both during walking and in static conditions. Peak pressures are defined as the maximum pressure values recorded for each sensor and are expressed in kPa. The resulting image of the foot is used to derive information of clinical value [4].

Baropodometric platform used: EPS+R it's the new generation of Loran Engineering resistive platform used to execute static and dynamic analysis of the foot. It can be used for postural analysis. EPS+R is usefull to transport and less influenced from the external environment. Get power data visualisation and accurate analysis in Biomech ® software [5].

#### WIVA motion sensors

An instrument called Wiva Science was used in this study. The Wiva Science is a motion analysis device based on the use of inertial sensors and the wireless transmission via Bleutooth of the acquired data to the associated smartphone, which is also made visible in real time on the PC. The sensor is positioned via a specific elastic belt on the patient's lower back at the level of the L4-L5 spinal vertebrae [6].

#### Protocol time up and go

The patient was assessed during walking with the use of motion sensors. The sensors were placed in the lumbar position on L4 and the Time Up And Go protocol was used [7,8]. The test consists of timing how long it takes a person to rise from a chair, walk 3 m, turn, and then sit again. In our protocol, the distance was increased to 6 m and performed twice. The TUG can be used to predict the risk of falls. A time of 35 s or more predicts falls with a likelihood ratio of 2.6 [9]; completing the test in less than 15 s predicts a reduced risk of falls with a likelihood ratio of 0.1 [9].

#### Follow-up and outcomes

The values for the patient with Strumpell-Lorrain disease are provided for both the right and left sides of the body, while the values for healthy individuals represent an approximate average range considered normal for the specified parameters.

#### Discussion

This clinical case contributes significantly to the field of podology by providing a detailed understanding of the kinematic challenges faced by patients with Strumpell-Lorrain disease. Strumpell-Lorrain disease is a neurodegenerative genetic condition characterized by progressive muscle weakness, spasticity in the lower limbs, and abnormal gait, resulting in difficulties in walking and maintaining balance [12]. The static analysis, depicted in Fig. 1, reveals a striking forefoot load predominance when the patient is in a static position. This phenomenon can be attributed to the spasticity affecting the Achilles tendon, resulting in restricted ankle dorsiflexion and an initial equinus foot position. It is noteworthy that despite this restriction, the patient retains some degree of active dorsiflexion, indicating an incomplete limitation [13]. Our assessment of plantar pressures during dynamic situations, as illustrated in Fig. 2, provides further insights into the patient's condition. The graph highlights elevated pressure levels at the left metatarsal area, with higher maximum pressure observed on the right side, accompanied by greater velocity on the right as well. These findings corroborate the presence of an equinus deformity, particularly pronounced on the left side, which elevates the vulnerability of the metatarsals to irritation or injury [14]. Further insights are derived from the force curves in Fig. 2, indicating a reduced emphasis on heel strike during the initial phase of gait, a common occurrence in early equinus foot. Conversely, the propulsive phase of gait is more prominent. The spatiotemporal parameters listed in Table 1 underscore the patient's challenges in gait efficiency and postural control, including slower walking speed, less efficient gait, and an altered gait cycle, all contributing to difficulties in maintaining balance and weight-bearing on one foot [15]. This comprehensive analysis offers valuable insights into the specific obstacles faced by individuals with Strumpell-Lorrain disease, particularly during the swing phase of walking and ankle dorsiflexion issues caused by spasticity in the gastrocnemius muscle. These findings provide a solid foundation for the development of tailored rehabilitation interventions. These interventions may include exercises designed to elongate the posterior kinetic chain and enhance balance during monopodal positions. The significance of this research extends to its potential to significantly improve the quality of life for individuals afflicted by this debilitating disease [16]. This clinical case study offers substantial insights into the kinematic challenges and biomechanical abnormalities associated with Strumpell-Lorrain disease. By leveraging advanced technologies like baropodometric footboards and motion sensors, we have conducted a comprehensive evaluation of the patient's gait characteristics, foot mechanics, and pressure distribution patterns. These findings highlight the presence of equinus deformity, forefoot load predominance, and altered gait parameters, deepening our understanding of the specific hurdles faced by Strumpell-Lorrain disease patients, particularly in walking, ankle dorsiflexion, and balance maintenance [17]. The implications of this research transcend this individual case, providing a basis for the development of targeted rehabilitation strategies. These strategies aim to address the mobility limitations and enhance the functional independence of individuals living with Strumpell-Lorrain disease. Moreover, this study underscores the immense potential of advanced technologies in the realm of podology. The objective assessment and continuous monitoring of neurodegenerative disorders' impact on gait and foot function through motion sensors and baropodometric platforms offer invaluable data for personalized treatment strategies. This, in turn, contributes significantly to the overall enhancement of patients' quality of life [18].

In conclusion, this clinical case provides valuable insights into the kinematic challenges faced by individuals with Strumpell-Lorrain disease, emphasizing the importance of tailored rehabilitation interventions. Further research in this area has the potential to enhance our understanding of the disease and improve the management and care of affected individuals.

#### Availability of supporting data

The protocol and the dataset analyzed during the current study is available from the corresponding author on reasonable request.

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#### **Declaration of Competing Interest**

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

#### References

- R. Schüle, L. Schöls, Genetics of hereditary spastic paraplegias, Semin Neurol. 31 (5) (2011) 484–493.
- [2] J.K. Fink, Hereditary spastic paraplegia, Neurol. Clin. 20 (3) (2002) 711–726.
- [3] D. C, S. G, B. A, D A, Hereditary spastic paraplegias: an update, Curr. Opin. Neurol. 20 (6) (2007) [Internet]Dec [cited 2023 May 31]Available from, https://pubmed. ncbi.nlm.nih.gov/17992088/.
- [4] R. Tedeschi, F. Giorgi, Manual therapy in diabetic patients with tibio-tarsal dorsiflexion deficit and forefoot overload: a case report, Int. J. Motor Control Learn. 4 (3) (2022) 0. Aug 10–0.
- [5] Loran Engineering science and technology of the movement for research, for medicine and for sports [Internet]. [cited 2022 Jul 2]. Available from: http://www. loran-engineering.com/baropodometric\_plate.html.
- [6] Wiva inertial system for performance analysis [Internet]. [cited 2022 May 8]. Available from: http://www.e-wiva.com/.
- [7] W. Browne, B.K.R. Nair, The timed up and go test, Med. J. Aust. 210 (1) (2019) 13–14.
- [8] Timed up and go test (tug) temporal phases assessment using a wireless device (free4act®): method validation in healthy subjects - ScienceDirect [Internet]. [cited 2022 Jul 2]. Available from: https://www-sciencedirect-com.ezproxy.unibo. it/science/article/pii/S0966636213000830?via%3Dihub.

- [9] E. Nordin, N. Lindelöf, E. Rosendahl, J. Jensen, L. Lundin-Olsson, Prognostic validity of the timed up-and-go test, a modified get-up-and-go test, staff's global judgement and fall history in evaluating fall risk in residential care facilities, Age Ageing 37 (4) (2008) 442–448.
- [10] B. Rw, Comfortable and maximum walking speed of adults aged 20-79 years: reference values and determinants, Age Ageing 26 (1) (1997) [Internet].Jan [cited 2023 Jun 10]Available from, https://pubmed.ncbi.nlm.nih.gov/9143432/.
- [11] M. Hb, L. Md, T. A, M.S.K. M, Lord Sr, Reliability of the GAITRite walkway system for the quantification of temporo-spatial parameters of gait in young and older people, Gait Posture 20 (1) (2004) [Internet]Aug [cited 2023 Jun 10]Available from, https://pubmed.ncbi.nlm.nih.gov/15196515/.
- [12] O. Walusinski, A historical approach to hereditary spastic paraplegia, Rev. Neurol. (Paris) 176 (4) (2020) 225–234.
- [13] W.K. Lam, J.C.Y. Leong, Y.H. Li, Y. Hu, W.W. Lu, Biomechanical and electromyographic evaluation of ankle foot orthosis and dynamic ankle foot orthosis in spastic cerebral palsy, Gait Posture 22 (3) (2005) 189–197.
- [14] B.E. Bloks, L.M. Wilders, J.W.K. Louwerens, A.C. Geurts, J. Nonnekes, N.L. W. Keijsers, Quantitative assessment of plantar pressure patterns in relation to foot deformities in people with hereditary motor and sensory neuropathies, J. Neuroeng. Rehabil. 20 (1) (2023) 65.
- [15] M. Manca, G. Ferraresi, M. Cosma, L. Cavazzuti, M. Morelli, M.G. Benedetti, Gait patterns in hemiplegic patients with equinus foot deformity, Biomed. Res. Int. 2014 (2014), 939316.
- [16] I. Campanini, M.C. Bò, F. Salsi, M.C. Bassi, B. Damiano, S. Scaltriti, et al., Physical therapy interventions for the correction of equinus foot deformity in post-stroke patients with triceps spasticity: a scoping review, Front. Neurol. 13 (2022), 1026850.
- [17] C. Buckley, L. Alcock, R. McArdle, R.Z.U. Rehman, S. Del Din, C. Mazzà, et al., The role of movement analysis in diagnosing and monitoring neurodegenerative conditions: insights from gait and postural control, Brain Sci. 9 (2) (2019) 34.
- [18] A. Arntz, F. Weber, M. Handgraaf, K. Lällä, K. Korniloff, K.P. Murtonen, et al., Technologies in home-based digital rehabilitation: scoping review, JMIR Rehabil. Assist. Technol. 10 (2023) e43615.