Case report

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Gnathological and osteopathic treatments with digital evaluations before and after therapies: a case report of a patient with ehlers-danlos syndrome

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Abstract

Background. Ehlers-Danlos syndromes (EDS) are inherited connective tissue disorders, requiring specific care along with a multidisciplinary approach by an expert medical staff, as for both the diagnosis and the treatment management. Chronic generalized pain and musculoskeletal dysfunctions due to joint hypermobility are common traits. Temporomandibular disorders (TMDs) are among the most frequent complaints. The authors report the diagnostic and therapeutic management of an EDS subject.

Case presentation. A 33-year old woman with Ehlers-Danlos syndrome, hypermobile type, was clinically evaluated and treated. She underwent a gnathological therapy with an anterior repositioning splint and proprioception exercises, and a physical rehabilitation through an osteopathic manipulation treatment (OMT). Technology – surface Electromyography (sEMG) of masticatory muscles, T-scan and stabilometric platform- supported diagnostic and therapeutic phases, giving objective and quantifiable information on the patient's assessment before and after treatments.

Conclusion. According to a targeted therapeutic strategy, both the gnathological and the osteopathic therapy proved to be effective in improving patient's symptomatology and functionality, as confirmed by digital findings. ***Clin Ter 2021; 172 (3):179-185. doi: 10.7417/CT.2021.2309***

Key words: *Ehlers-Danlos syndromes, gnathological therapy, osteopathic manipulation treatment, sEMG, T-scan, stabilometric platform, case report*

Background

Ehlers-Danlos syndromes (EDS) are a group of hereditary systemic pathologies, affecting the connective tissue, characterized by hypermobility of joints, hyperextensible skin and musculoskeletal, visceral, pelvic and neurologic dysfunctions [1]. The variety of clinical aspects of such disorders requires care along with a multidisciplinary approach. Dental practitioners play an important role in screening and treatment [2]. Severe temporomandibular pain and dysfunctions are common traits in the Ehlers-Danlos syndromes [3]. This is in all likelihood connected with hypermobility, frequency of subluxations and dislocations, soft tissue injury and myalgia. Furthermore, pain and dysfunctions interfere with patients daily-life, triggering psychological issues.

Multiple treatments are available for management of pain and TMD. Among conservative treatments, the splint therapy strengthened by proprioception exercises might be an option as supportive treatment [2]. The therapeutic goals of the gnathological therapy are the reduction of joint load, the muscle rest and the normalization of mandibular movements [4].

Chronic, early onset, debilitating general musculoskeletal pain is usually associated to EDS, and in particular to the hypermobile type. This is due to the repeated traumas during episodes of subluxation and uncontrolled movements and the presence of joint instability and impairment in muscular functionality. External factors may have an impact as well, such as lifestyle, sport activities, surgery and other comorbidities. Several treatment strategies may be used to face generalized body pain such as manual therapies, the cognitive-behavioral therapy and pharmacologic treatments. Among manual therapies, osteopathic manipulation therapy (OMT) is an option [5,6]. This kind of physical therapy aims at restoring the structural and functional integrity of the organism, when altered by somatic dysfunctions, and at stimulating its intrinsic tendency to self-healing, through a wide range of manual therapeutic techniques. A structural diagnosis is an essential component of osteopathy [7].

Clinical and radiographic evaluations during the diagnosis and after the therapy remain the gold standard, however

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the introduction of digital technology allowed to "measure", quantitatively and qualitatively, the evaluated data. In this case-report, the digital devices supported the diagnosis and the evaluation of the results obtained with different kind of treatments [8].

In line with such considerations, a clinical case of an EDS patient treated with occlusal splint, mandibular proprioception exercises and OMT, evaluated before and after treatments with Surface Electromyography (sEMG) of masticatory muscles, T-scan for the analysis of dental occlusion and stabilometric platform was reported.

Case presentation

The study was approved by the Institutional Ethics Committee of Department of Oral and Maxillo-Facial Sciences, Sapienza University of Rome (PROTOCOL NO. 0001385) and the patient signed the informed consent, according to the World Medical Association's Declaration of Helsinki. The study was conducted between June 2018 and December 2019.

A 33-year old woman with temporomandibular disorders and affected by Ehlers-Danlos syndrome (hypermobile type -hEDS) was evaluated by a specialized medical staff at the Department of Oral and Maxillo-facial Sciences, Policlinico Umberto I, Sapienza University of Rome, after an initial specialistic medical examination at the Department of Rare Diseases, Policlinico Umberto I, Sapienza University of Rome.

She had an history of moderate pain - Verbal Numeric Scale (VNS) = 50 - at the level of the left Temporomandibular joint and muscular pain (VNS= 50) at the level of the left masseter. She reported about 5 episodes/week of bilateral temporal and occipital headache (VNS =70) and bilateral neck pain of mild intensity (VNS=20); neither tinnitus nor dizziness.

The patient also reported general musculoskeletal pain: brachialgia (VNS right 40 -left 20); dorsal pain (right 40-left 40); lumbar pain (right 70- left 50); sacroiliac joint pain (right 50-left 40); pain due to femoropatellar chondropathy (right 50-left 40).

At the clinical intra-oral examination, she showed Class I dental occlusion, no occlusal anomalies, signs of tooth wear, normal overjet and overbite. At the gnathological examination, she showed bilateral temporomandibular joints subluxations and pain on the left Temporomandibular joint (TMJ) (point 1-2 Rocabado pain map), tenderness at masseters, lateral pterygoids and anterior/medium/posterior bundles of temporalis muscles, excessive mouth opening excursion (= 52mm), no latero-deflection in mandibular opening movement. The Magnetic Resonance Imaging showed a normal position of TMJs discs but bilateral flattening of temporal glenoid fossa.

The patient also underwent a specialistic visit by an osteopathic physician. Diagnosis was based on the five bioenergetic models [5] and in accordance with the integration of osteopathic principles, the identification of underlying etiologies and a proper osteopathic differential diagnosis by considering somatic dysfunctions [9].

From the collection of the anamnestic data, the osteopathic physician reported the specific information about each system and structure in accordance with the five bioenergetic models [5] in a summarizing table, reported below- Figure 1. Furthermore the patient's basic postural pattern was considered. The patient presented with an external rotation of the right half body and the internal rotation of the left half body; right lower limb longer than left one with compensation by internal rotation.

Biomechanics	Neurologic	Respiratory-circulatory	Metabolic	Behaviour
- Complicated birth, with caeserean section - Osteopenia and cervical/ lumbar disc protrusions - Ehlers-Danlos syndrome hypermobile type	- Myopia	- History of tonsillitis and subsequent tonsillectomy - Mitral valve insufficiency - 1 episode of pneumonia ab gastro-esophageal reflux - ovarian cysts, vulvodynia, pudendal neuralgia	- Food intolerance (lactose) - Hiatal hernia - Intestinal disease	- Mild level of somatization

Fig. 1. Anamnesis based on the five models of osteopathic synthesis

After these clinical examinations, the patient underwent three in-depth digital tests: surface Electromyography of masticatory muscles (TMJOINT, BTS SpA, Garbagnate Milanese, Italy), T-scan III (produced by Tek-Scan, Boston USA) for the analysis of dental occlusion and Stabilometric platform (DIASU, by Sani Corporate, Rome Italy) for the posture evaluation.

The sEMG evaluation followed such assessment criteria. The authors of the study used the standardized protocol proposed by Ferrario [10] . This protocol allows to calculate indices of jaw muscles activity by using standardized EMG signals recorded during the maximum voluntary contraction in maximal intercuspation and on cotton rolls.

The electrical activity of the right and left anterior bundle of Temporalis Muscles (TA) and right and left masseters (MM) was recorded simultaneously during standardized tasks.

The preliminary analysis before treatment led to the following results.

BAR (muscle barycenter) index, referring to the stomatognathic muscular center of gravity, had an anterior right position indicating the prevalence of temporalis muscles on masseters. Furthermore, IMP (Impact) index referring to muscular strength during clenching was abnormally increased, revealing an excessive muscular work due to parafunctions. There were no alterations in symmetry indexes (POC MM and TA- percentage of overlapping coefficient for masseters and anterior temporalis; ASIM- asymmetry index- between the left and right sides; TORS- torque coefficient).

As for the analysis of dental occlusion, T-scan test was performed, according to the protocol under the study of Ferrato et al [11].

Before treatments analysis reported the following outcome.

The occlusal center of gravity was on the right side. It was placed before the second molars, that is the specific occlusal fulcrum of the patient under consideration, and it indicated an "abnormal thrust" of the occlusal loads in the anterior direction. Furthermore, the black line, indicating the occlusal strength over time, showed a moderate loss of neuromuscular efficiency.

The postural and stabilometric evaluation was conducted using a stabilometric platform, recording the balance conditions through the dynamics of the center of gravity, and the postural assessment through the distribution of the human body loads, with the analysis of the plantar support.

The patient was evaluated during her quiet standing, with her feet apart at approximately 30° and a distance of about 5-6 cm, while observing a fixed point in front of her at a distance of between 60 cm and 1mt. The standardized, repeatable and reproducible indices considered for an homogenous diagnostics of the perturbations of the postural balance were: the index assessing the visual interference (Romberg index)[12] and the indices evaluating the stomatognathic interference and the cervical interference (according to Guidetti's protocol)[13].

Digital data allowed to detect the presence of interference of the oculomotor system as shown by an increased surface of the ellipse when the patient's eyes were open and a significant impairment of the cervical spine as shown by the alteration of both the surface of the ellipse and the length of sway when the patient's head was retroflexed. No stomatognathic interference on postural balance was shown. Furthermore, the body load on plantar support was higher on the right side with a bilateral prevalence on the hindfoot.

Once the diagnostic procedure had been completed, on the basis of the specific diagnosis, the patient underwent a gnathological therapy for the treatment of the stomatognathic component, and osteopathic manipulation treatment (OMT) for the management of pain and dysfunctions of the spine and the other musculoskeletal disorders [14]. Furthermore, the patient underwent a specialist eye examination.

Gnathological therapy consisted of the use of a direct tridimensional repositioning appliance (DI.TRA) [4], in compliance with the gnathological protocol of our School whose effectiveness and efficiency have been already evaluated in a recent clinical study [4]. The therapy with the occlusal splint was accompanied by proprioception exercises. Proprioception exercises consisted of isometric tongue-strengthening exercises, carried out positioning the tip of the tongue on the hard palate behind the upper incisors, in order to improve swallowing, muscular function and articular coordination [14]. The duration of gnathological therapy was 18 months.

As for the osteopathic treatment, the most recent osteopathic strategies for the treatment of chronic pain and musculoskeletal dysfunctions were applied [14,16]. These strategies aim at addressing body unity issues, enhancing homeostatic mechanisms and maximizing structure-function inter-relationship.

The patient underwent three cycles of OMT, two months apart from each other, each cycle consisting of four sessions (once session a week, every week for a month). Each treatment lasted 45 minutes.

The synthesis of these cycles is reported in the table below – Figure 2.

For the purpose of this study, only the treatment of the musculoskeletal dysfunctions and pain is reported, although the patient was also treated for the visceral dysfunctions, found during the medical history and during the first osteopathic evaluation.

VNS, clinical and digital examinations were performed after the end of therapies and they showed the following outcome.

Neither temporomandibular joint pain nor masticatory muscle pain were reported. The temporal and occipital headache was reduced in both intensity and frequency (VNS from 70 to 40). No neck pain was reported. The patient gained a greater control of mandibular movements through proprioception exercises and underwent a real "cognitive behavioral therapy" to tackle the habit to clench and grind.

After the OMT the patient referred: brachial and dorsal pain of mild intensity (VNS right/left 10); a significant decrease of lumbar and sacroiliac joints pain (VNS right 30-left 20); an improvement of pain due to femoropatellar chondropathy (VNS right/left 10).

As far as the sEMG scores are concerned, the values fell within the reference ranges. Changes in the BAR index indicated an improvement in the muscular balance and a better stability. The "normalization" of IMP value showed a decrease in the rate of clenching and muscles work.

T-scan analysis showed that the occlusal center of gravity fell within a normal range in latero-lateral direction and re-

Session 1	Session 2	Session 3	Session 4
FIRST CYCLE			
- right iliac outflare - left Temporomandibular joint pain - tenderness at masseters, temporalis and pterygoid muscles	- osteopathic reassessment - stabilization of previous cycle results	- osteopathic reassessment - stabilization of previous cycle results - lumbar spine dysfunction - cervical spine dysfunction	- osteopathic reassessment - stabilization of previous cycle results - shoulder and thoracic girdles dysfunction
SECOND CYCLE			
- osteopathic reassessment - stabilization of previous cycle results - right iliac outflare	- osteopathic reassessment - stabilization of previous cycle results - cervical and lumbar spine dysfunction - skull	- osteopathic reassessment - stabilization of previous cycle results	- osteopathic reassessment - stabilization of previous cycle results
THIRD CYCLE			
- osteopathic reassessment - stabilization of previous cycle results - cervical and lumbar spine dysfunction - skull	- osteopathic reassessment - stabilization of previous cycle results - sacrum	- osteopathic reassessment - stabilization of previous cycle results	- osteopathic reassessment - stabilization of previous cycle results - dorsal spine pain

Fig. 2. Dysfunctions detected and treated during each cycle of Osteopathic manipulation treatment.

gained a better position in posteroanterior direction (closer to the Maximum Occlusal Load -MOL). Furthermore, the black line showed a better neuro-muscular stability.

Changes in the Romberg index were recorded, due to an improvement in the oculomotor system, after visual rehabilitation. Also, the cervical parameters showed an improvement, in particular of the sway length ratio when the patient's head was retroflexed, but also of the ellipse surface, resulting in an improvement of the "stability" of the body center of gravity and its projection on plantar support. A significant load redistribution between right and left foot and between forefoot and hindfoot was shown.

Digital evaluation outcome before and after treatment is reported in the Figure 3.

The timeline of the diagnostic and therapeutic phases is reported in the Figure 4.

Discussion and conclusions

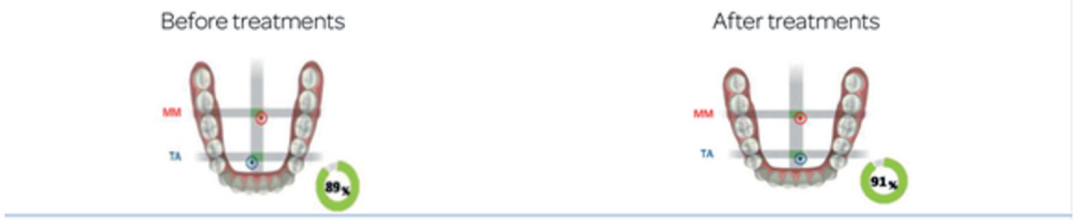
In the scientific literature there are only few studies on the management of TMD patients with EDS. As for the gnathological approach, it is worth mentioning the study of Mitakides et al [2], who proposed several types of treatments such as deep heat for muscles relaxation, cold laser; muscular massages; regenerative injection therapy for ligament and tendon regeneration; medications; botulinum toxin; physical therapy and surgical options for damaged TMJs. Also splint therapy proved to be effective and it is indeed the therapeutic strategy adopted in this study.

The gnathological protocol used for this project is compliant with the guidelines by our gnathological School on the choice and management of occlusal splints in the treatment of TMDs [7], specifically tailored for this medical condition. Splint was applied to give appropriate anterior repositioning, provide articular stabilization, limit joint injury, as well as to help maintaining physiological posture [17]. The results obtained include the withdrawal of painful symptomatology at the level of temporomandibular joints and related masticatory muscles and the drastic reduction of the headache, being the major complaint of the patient, among cranio-facial aches. The reason for combining gnathological therapy using occlusal splints with exercises to improve proprioception derives from the instability of EDS clinical condition. Proprioception exercises as "cognitive-behavioral therapy" should always be carried out in combination with a splint treatment to stabilize the achieved results, ensuring a better and more lasting stability. In fact, loss of proprioception in EDS has been reported in the medical literature and it is considered to be an important factor in EDS-related chronic pain [18]. Exercises, aimed at improving proprioception, proved to reduce pain. Improvement of proprioception might be effective to ameliorate both the functional status and the chronic pain.

In fact, in TMD patients, occlusal splints should not be applied for a life-time but only for a limited span of time,

A

EMG index	Before treatment Value (%)	After treatment Value (%)	Reference range
POC TA	88.66%	90.14%	83 ≤ (%) ≤ 100
POC MM	88.09%	89.81%	83 ≤ (%) ≤ 100
BAR	**89.53%**	91.10%	90 ≤ (%) ≤ 100
TORS	91.52%	93.14%	90 ≤ (%) ≤ 100
IMP	**395.47%**	99.30%	85 ≤ (%) ≤ 115
ASIM	0.72%	-1.15%	-10 ≤ (%) ≤ 10



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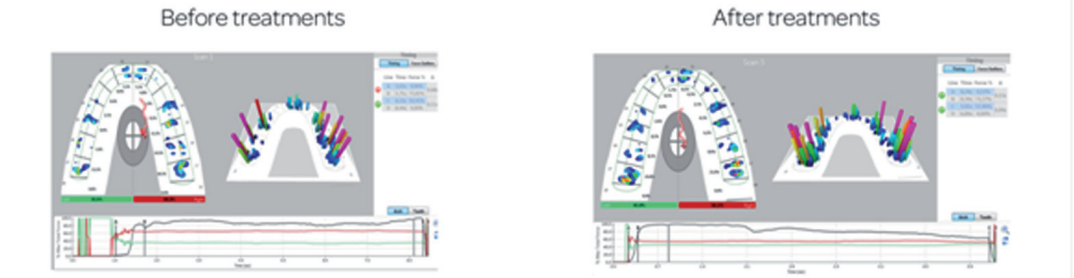

C

VISUAL INTERFERENCE: ROMBERG INDEX VALUE <100 IS CONSIDERED SIGNIFICANT) VALUE <100 IS CONSIDERED SIGNIFICANT)	BEFORE TREATMENT **ES CE/ ES OE = 20.66** SL CE/ SL OE = 102.83	AFTER TREATMENT ES CE/ ES OE = 120.0373 SL CE/ SL OE = 123.2309
STOMATOGNATHIC INTERFERENCE Value < 60 is considered significant Value <70 is considered significant	ES CE OR /ES OE OR = 85.68 SL CE OR/ SL OE OR = 91.27	ES CE OR /ES OE OR = 161.99 SL CE OR/ SL OE OR = 102.75
CERVICAL INTERFERENCE Value >120 is considered significant Value >120 is considered significant	**ES CE RH/ ES OE RH = 307.98** **SL CE RH/ SL OE RH = 128.17**	**ES CE RH/ ES OE RH = 139.75** SL CE RH/ SL OE RH = 118.85

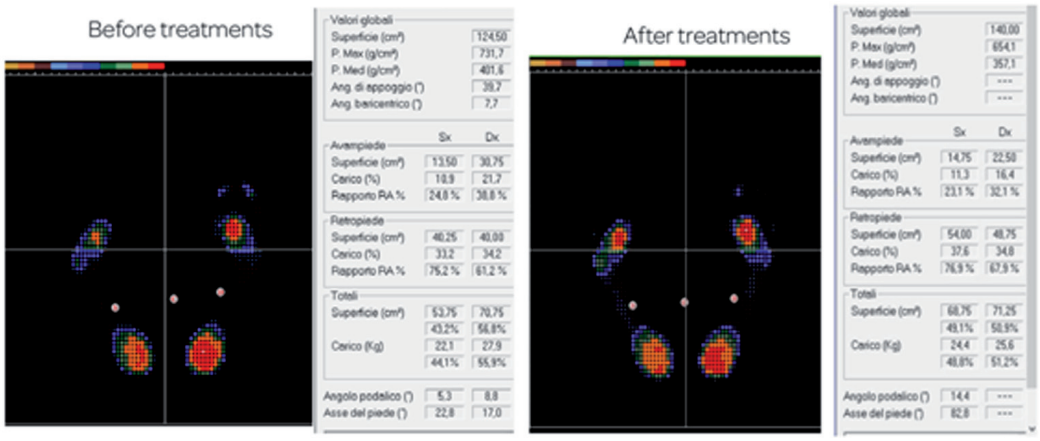

Fig. 3. A. sEMG values compared to reference ones before and after treatments. B. Position of the occlusal center of gravity and occlusal forces expressed in percentage before and after treatments. C. Stabilometric evaluation indexes, distribution of the center of pressure and evaluation of plantar support before and after treatments (ES (Ellipse surface), CE (closed eyes), OE (open eyes), SL (sway length), OR (occlusal release); RH (retroflexed head).
Bold type= out of reference range.

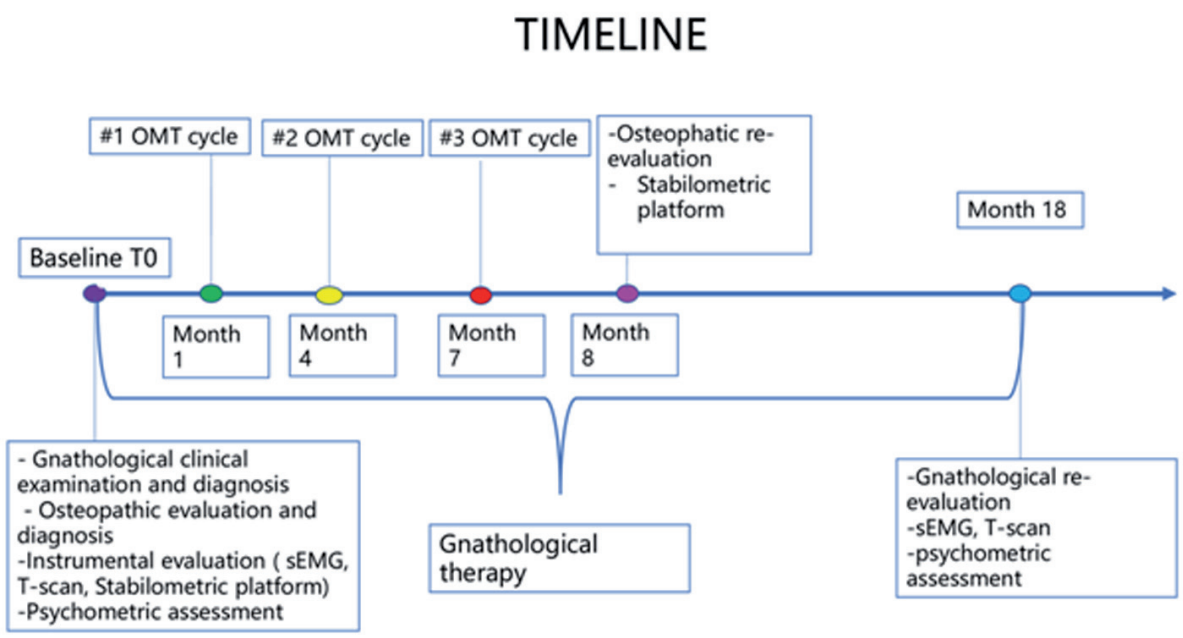

Fig. 4. Timeline of the diagnostic and therapeutic phases.

long enough to restore TMJs and related structures. However, in EDS patients, recurring instability and several risk factors due to the syndrome concern the clinicians upon suspension of therapy. Therefore ongoing constant follow-ups are certainly mandatory.

Gnathological digital devices are not yet eligible as diagnostic tools and do not directly link the "digital signs" neither with the clinical one nor with the symptoms, as other equipment do, such as electrocardiogram. However, the surface electromyography certainly helps in getting knowledge of the characteristics of the muscular pattern of this type of patients, otherwise not quantifiable by clinicians. As for sEMG evaluation, the index (BAR) is a good indicator of stomatognathic dysfunction because it is linked to a reduced masticatory efficiency in the posterior areas of dental arches, often found in a condition of mandibular retropositioning or reduced vertical dimension. The IMP index also allowed to give a quantifiable value of the parafunctional muscular activity. Previous studies used sEMG to evaluate the clinical outcome of gnathological therapy, showing the ability of such digital device to detect changes in muscular activity over time, after orthopedic therapy with occlusal splints [8]. Under such circumstances, the positive change of sEMG pattern drags along the one of clinical signs and symptoms. In this study a normalization of parameters was recorded, in line with the study of Tecco et al [8].

The digital results obtained by the T-scan are in line with the ones obtained with the electromyography. The only difference is that this device "observes" the system from another point of view, that is precisely the occlusal one. The occlusal component is often the "victim" of an alteration of the stomatognathic system and this is proved by the fact that without permanent alterations of the occlusion, by only restoring joint and muscular components, also the occlusal loads were more evenly distributed [11].

Chronic pain is one of the major symptoms presented by patients with EDS [19]. Pain management strategies should therefore focus on the etiology of pain and reduce pain sensation. As for the treatment of the musculoskeletal pain and dysfunctions, the evidence suggests that patients undergone physical treatments generally improve over time [20]. Physical rehabilitation consists of core stabilizing and joint stabilizing [21,22]. Stretching exercises should be limited to gentle stretching to avoid any risks of subluxations or dislocations. Techniques that have been used in treating EDS pain include manual therapy for overactive muscles, trunk stabilization, posture re-education, joint awareness using biofeedback, joint mobilization with muscle release [21].

Often a holistic approach such as the osteopathic one is integrated in the multidisciplinary treatment. Based on its bioenergetic models [5], osteopathic manipulative treatment helps to induce articular release resulting in increased joint motion, reduced pain as well as improved blood flow, lymphatic drainage, and proprioception. In light of the increased tissue fragility and weakness of supporting structures of the joints, gentler techniques allowing positional release and counter-strain are highly recommended [14-16] . Usually a combination of models of treatment is applied to each patient. The chosen combination is modified according to the patient's differential diagnosis, comorbidities and other treatment regimens.

The patient examined in this study had a good degree of improvement after OMT cycles. The good results obtained have also been verified by the positive trend of the stabilometric platform indices, which showed a better and more balanced distribution of the body loads between right and

left foot and an improvement of the body center of gravity. The improvement of the cervical interference index, as proved by our clinical experience, actually refers not only to an improvement of the cervical tract but most likely of the spine as a whole.

Multidisciplinary approach proved to be an effective strategy for the management of the EDS patient. Well-trained medical staff and tailored treatments are mandatory, when treating this complex medical condition. Digital devices supported the diagnosis and the therapeutic phase, giving quantitative information on the patient's assessment.

Declarations

Availability of data and materials. All data sets are included in the manuscript.

Competing interests. The authors declare that they have no competing interests.

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The study was carried out according to CARE checklist.

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