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VETOSED study: efficacy of compressive garments for patients with hypermobile Ehlers–Danlos syndrome on shoulder stability and muscles strength

Emmanuelle Chaléat-Valayer\textsuperscript{a}, Angélique Denis\textsuperscript{b}, Amélie Zelmar\textsuperscript{b}, Anne Pujol\textsuperscript{a}, Amandine Bernadou\textsuperscript{a}, Rachel Bard-Pondarré\textsuperscript{a} and Sandrine Touzet\textsuperscript{b}

\textsuperscript{a}Centre Médico-Chirurgical de réadaptation des Massues, Croix-Rouge française, Lyon, France; \textsuperscript{b}Pôle de Santé Publique, Hospices Civils de Lyon, Lyon, France

ABSTRACT

Purpose: to assess the effect of wearing a compressive short-sleeve jacket on shoulder stability and rotator muscles in adult patients with a hypermobile subtype of Ehlers–Danlos syndrome.

Materials and methods: a quasi-experimental study with “Pre/Post” design (4 weeks with versus 4 weeks without), open, conducted on a national cohort. Shoulder rotators were assessed with an isokinetic device at 180°/s and 90°/s; frequency of shoulder stability defects, pain (Visual Analogical Scale), and satisfaction (Quebec User Evaluation of Satisfaction with Assistive Technology) were reported.

Results: 36 patients (35 women), mean age 37.9 years, wore jackets for a meanly 7.8 h/day. >70% were satisfied with the effect on arthralgia, instability, or function. QUEST results were high (m = 4.1, SD = 0.7). After jacket wear, the power of external rotators at 180°/s was significantly increased (+1.29 W, CI95%: 0.31; 2.28; \( p = 0.0318 \)). At 90°/s, no significant difference was found, either on power or strength. The on-off effect highlights a significant difference in external and internal rotators power, whatever the speed. The occurrence of subluxation (\( p = 0.0140 \)) and dislocations (\( p = 0.0163 \)) decreased. Pain decreased from 3.5/10 to 2.5/10, without significance (\( p = 0.0964 \)).

Conclusion: compressive CICATREX SED\textsuperscript{c} jackets are well supported by patients, impact the power of external rotators at high speed (180°/s), and improve joint stability.

IMPLICATIONS FOR REHABILITATION

- Compressive garments made to measure are beneficial to patients with hypermobile Ehlers–Danlos syndrome.
- Compressive CICATREX SED\textsuperscript{c} jackets clearly improve shoulder stability and help to promote muscle power in shoulder external rotators during high-speed movements.
- One month of jacket wearing seems to bring no lasting effect on shoulder-rotator power, so the jacket needs to be kept on for the benefits to be maintained.

INTRODUCTION

Ehlers–Danlos syndrome (EDS) is a rare genetic disease associated with connective tissue disorder [1,2]. This rare syndrome affects mainly women, and its prevalence is generally estimated between 1/5000 to 1/10 000 [3]. Several subtypes are identified [4,5]; the most frequent EDS subtypes are the classical and hypermobile subtypes (hEDS), the two of them representing 80%. The hEDS is characterized by hypermobility and generalized joint hyper laxity, accompanied by hyperextensible, velvety, and thin skin [5]. Joint hyper laxity is due to hyperelasticity of the ligaments but also of the tendons and muscles. It causes poor joint centering and imbalance between the agonist and antagonist muscles [6]. Hyper laxity thus leads to joint hypermobility [6] characterized by excessive joint flexibility that can be demonstrated by a Beighton score greater than 5 [7]. The hEDS is manifested by multiple joint dislocations and subluxations, as well as repeated sprains, occurring either spontaneously or secondary to minimal trauma. This context leads patients to an attitude of avoidance of potentially traumatic situations, through a phenomenon of painful anticipation, resulting in muscle deconditioning and passive maintenance of hyperlaxity not compensated by muscle mass that has become inactive. Pain is present in most of these patients [8]. These are mainly joint pains (arthralgia) that are intermittent or continuous and can affect all joints. They are most often mechanical (maximum under stress), increase with movement, and require frequent position change [6,9]. Fatigue and misadaptation to the effort are frequent and disabling [10]. They are manifested by asthenia and muscle discomfort in addition to periarticular pain and reflex protection of joint instabilities to limit functional abilities [11].

Among the therapeutic options [9,12], compressive garments appear as medical devices designed specifically for hEDS in the symptomatic treatment of joint pain, in order to get a functional improvement by stabilizing joints through a proprioceptive effect [13] and some muscle globulation [14]. Unfortunately, very few studies explore the effect of compressive garments in hEDS [14,15] and they bring insufficient evidence for their effect
because of the methodology used (lack of control group and primary outcome measure).

The aim of this study, called VETCOSED (VETements COMpressifs pour le syndrome d’Ehlers Danlos), was to evaluate if wearing a compressive garment in patients with hEDS would decrease the painful symptomatology and improve joint stability, by changing the muscular balance, in the shoulder area, as patients with hEDS are at particular risk of dislocation and pain in this joint.

**Material and methods**

**Design**

This study used a quasi-experimental design (wearing a compressive jacket versus no compressive jacket), open, conducted on a cohort of patients with hEDS, taken as their own control.

The study was national in scope, with patients recruited throughout France by an information campaign conducted by the French association of patients with EDS (AFSED), a press briefing via APICIL foundation (which is a French foundation entirely dedicated to pain, enabling the emergence of innovative and more humane programs to improve pain management), about twenty letters sent to doctors caring for patients with EDS (physicians in physical and rehabilitation medicine, geneticists, rheumatologists, specialists in internal medicine), and regular newsletters.

For all participants, evaluations were conducted always by the same evaluator at one specific rehabilitation center, a physiotherapist specialized in the isokinetic assessment and known as an expert for this pathology.

After signing the consent, included patients were equipped with a made to measure CICATREX SED® compressive jacket with short sleeves (Figure 1). Two visits were necessary, supervised by an occupational therapist: one for taking measurements of the patient and one for retouching the garment. Patients were then followed for 8 weeks, with 2 follow-up visits: after the 4 weeks with compressive jacket (day 28), and then after 4 weeks without compressive jacket (day 56) as shown by Figure 2. Due to the great heterogeneity of patients in terms of their history of wearing compressive garments, the data from the inclusion visit could not be considered as pre-intervention data.

In accordance with the medical prescription, the jacket had to be worn as much as needed, with a minimum of 3 h/day.

**Participants**

Patients had to present an hEDS syndrome confirmed by the clinical record of a geneticist, with hyperlaxity (Beighton score ≥5/9) [7], instability of one or both shoulders leading to pain (Visual Analogic Scale >0) or disability: recurrent dislocation or subluxation (at least once a month or at least 12 times a year). They had to be over 18-years-old, of a size equal to or greater than 1m40 due to technical aspects of the isokinetic assessment device. They had to be able to understand and answer questionnaires, benefitting from health insurance, agreeing to participate in the study and not subject to legal protection, and having signed their informed consent.

Patients were not included if they had an unstable cardiac pathology, a history of shoulder surgery (abutment), severe skin damage, or an ongoing hyperalgic episode or if they were pregnant.

**Main and secondary criteria**

The main criterion was the efficacy of wearing a short sleeve compressive jacket (CICATREX SED® model) compared to its absence in improving the power of shoulder rotators, measured using an isokinetic device (CYBEX NORM® type), in patients with hEDS.

The secondary criteria were the impact of wearing a short sleeve compressive jacket on pain in the shoulders and joint stability (decentration, subluxation, dislocation); and the compliance with wearing the jacket.

**Evaluation criteria**

The main evaluation criterion was the measure of power of shoulder rotators with the isokinetic assessment [16], made at day 28 and day 56 with and without a garment. To perform the different measurements, each patient was wearing a wrist protective orthosis to avoid pain and damages during the evaluation and has been placed in a modified Davies position is currently recommended in pathological contexts [17]: sitting position, arm abducted between 25° to 45° in the plane of the scapula. The range of motion for each patient was determined according to the possibilities of the more injured side and applied to the contralateral side.

The power (in watts (W)) of the shoulder rotators was measured by concentric isokinetic evaluation at a constant angular velocity of 180°/s and 90°/s on isokinetic device (type CYBEX NORM®), and the power ratio of the external and internal rotators of each shoulder at a constant angular velocity of 180°/s and 90°/s. The 180°/s power measurement best represents the functional situation of daily life, where shoulder stability must be managed at high movement speeds. The evaluation at the angular velocity...
of 90°/s is close to the usual isokinetic training speed for this pathology.

The choice of the concentric mode is deliberate for this evaluation, as the eccentric mode is much more constraining and more difficult for fragile and painful people; eccentric evaluations require a little longer learning and are sometimes distressing for the subject who does not necessarily control the movement he is opposing.

**The secondary evaluation criteria**

1. The strength of the external and internal rotators of each shoulder at 180°/s and 90°/s expressed by the torque peaks (in newton-meters (N m)), measured at day 28 and day 56.
2. The frequency of shoulder joint stability defects (decenteration, subluxation, dislocation) either self-reduced (by the patient himself or spontaneously), reduced by someone else (doctor, physiotherapist), or not reduced (permanent instability). This was assessed on day 28 and day 56.
3. The patients’ level of pain in the shoulders, expressed by the Visual Analogic Scale (VAS), was assessed at rest at day 28 and day 56.

**Others**

The SF-36 questionnaire was used to assess the quality of life at inclusion; this valid generic self-administered questionnaire includes 36 questions divided into 8 domains: physical mobility and performance, limitations in the activities of daily life, social integration, restrictions in usual occupations for physical or psychological reasons, psychological suffering, vitality and perceived health [18]. Scores range between 0 (lowest level of quality of life) and 100 (best level of quality of life).

Daily functional independence was assessed at inclusion in our group using the Health Assessment Questionnaire (HAQ) [19], where scores range between 0 (no difficulty) to 3 (no capacity), and a perceived fatigue score was also determined at inclusion with the Fatigue Impact Scale (FIS), which is a 40-item questionnaire assessing fatigue in its cognitive, physical and social dimensions. [20] The higher the score, the higher level of fatigue is noted, with a maximum of 160 points.

The QUEST self-questionnaire (Quebec User Evaluation of Satisfaction with Assistive Technology) [21] is a 12-items outcomes measure that was used to assess user satisfaction with their compressive jacket with 2 components: Device and Service. The 12 items are rated on a 5-points scale, range from 1 ("not satisfied at all") to 5 ("very satisfied"). The QUEST was used on Day 28. The compliance was also observed at study visits, either with the wearing of the compressive garment during the interventional phase where the daily duration of jacket-wearing was determined by patient report or with the non-wear of the garment during the control phase.

**Sample size**

The number of subjects required was estimated at 34 patients to demonstrate a 30% improvement in the power of the external shoulder rotators when patients wore the compression garment for 1 month compared to that measured in the absence of the garment (i.e., an improvement of 2 W for an average external rotator power of 6.9 W without the compression garment measured at a constant angular velocity of 180°/s) considering a power at 80%, an alpha risk at 5% and a deviation of the difference of 4.

**Statistical analysis**

Statistical analyses were carried out using the software SAS 9.4 (SAS Institute Inc., Cary, NC, USA). Patients with documented primary outcomes were analyzed. Means and standard deviation (SD), median and quartiles values when appropriate, were calculated for continuous variables, and frequencies with percentages for categorical variables.

The efficacy of the compressive jacket on the power of the external rotators at 180°/s was assessed using repeating measures analysis of variance to take the multilevel structure of the data into account with isokinetic measure at level 1 nested within shoulder at level 2 nested within the patient at level 3. The model was controlled for shoulder dominance and history of wearing compressive garments as fixed effects and for the patient as a random effect.

The same model was built for the power of the internal rotators and the power ratio of the external and internal rotators at 180°/s. The significance level was set to an alpha of 0.05 and the Holm–Bonferroni method was applied to correct for multiple comparisons. The effect of the compressive jacket was evaluated with the same approach for the power of the shoulder rotators at 90°/s, for the strength of the shoulder rotators (torque peaks of

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**Figure 2. General design of VETCOSED study.**
Results

Characteristics of participants

A total of 46 patients were included. Of the 46 patients, 10 (22%) were excluded from the efficacy analysis: Figure 3 shows the study flow chart.

35 women and 1 man were included, with a mean age of 37.9 ± 11.6 years (range: 19–62). 89% (32/36) were right-handed. Half of them were in paid employment, and their working time exceeded 60% for 28% (5/18) of them, exceeded 80% for 67% (12/18) of them, and was less than 40% for one of them. Patients had had symptoms for an average of 25 ± 13.2 years (range: 3–53), but the average length of time since diagnosis was only 7 ± 7.5 years (range: 0–27).

All patients included had unstable shoulders, but instability generally concerned other joints: the ankle (100%), the knee (91%), the wrist (86%), the spine (80%), the hip (77%), the elbow (54%), fingers at the MCP (74%), PIP (63%) and DIP (57%), or other (54%). 78% (28/36) were taking medication, including pain medication for 82% of them (23/25).

At inclusion, the results for the SF-36 questionnaire was a meanly 30.4 for the physical domain (SD = 5.9) and 38.2 for the mental domain (SD = 10). The FIS total score was meanly 100.9 (SD = 32), and the HAQ score was meanly 1.4 (SD = 0.5).

Regarding compressive garments at the time of inclusion, 9 (25%) patients were not novo: 3 patients were already wearing a short-sleeved vest, and 6 had used one in the past. 64% of patients wore sometimes another type of compressive garments, such as gloves (61%), ankle braces (65%), panty (39%), or a long-sleeved jacket (13%).

Regarding the isokinetic assessment, the results show that the mean power was 9.8 W (SD = 8.5) at 180°/s and 9.7 W (SD = 7.2) at 90°/s in external rotators and 20.9 W (SD = 20.2) at 180°/s and 19.8 W (SD = 19.8) at 90°/s in internal rotators.

Impact on the shoulder rotators

Effects of the compressive jacket after 4-weeks of wear on power and strength of rotators are presented in Table 1.

At 180°/s, the power of the shoulders external rotators was significantly increased by 1.29 W (CI95%: 0.31; 2.28) after 4 weeks of wearing a compressive jacket compared to the situation without wearing it (Holm–Bonferroni adjusted p-value = 0.0318). The power of internal rotators was increased by 2.3 W (CI95%: 0.22; 4.38), but the difference was slightly non-significant after Holm–Bonferroni correction (p-value = 0.0620). No significant effect was shown on the ratio of external/internal rotators powers.

Regarding the strength of internal and external rotators, there was no significant effect of wearing a compressive jacket for 4 weeks.

At 90°/s, no significant difference was found after 4 weeks of wearing a compressive jacket, either on power or muscle strength.

Immediate effects of jacket removal after 4 weeks of jacket wear are shown in Tables 2. There was a significant on-off effect
on external and internal rotators power, whatever the speed, but no effect again on the strength of these muscles.

**Impact on pain and joint stability**

Table 3 presents the impact of 4-week jacket wear on joint stability and pain. After 4 weeks of wearing the jacket, 72% of patients had at least one episode of joint instability compared to 92% after 4 weeks without wearing the jacket. The difference was significant (Holm–Bonferroni $p$-value = 0.0326). Wearing a jacket significantly decreased the occurrence of subluxation ($p = 0.0140$) but also of dislocations ($p = 0.0163$).

Pain decreased after 4-week jacket wear, from 3.5/10 on the Visual Analogical Scale to 2.5/10, but this difference was not significant ($p = 0.0964$).

**Discussion**

Patients included in VETCOSED often had different compressive garments, but until now, no study explored their effective wearing nor their effect. The first important conclusion of this study is that compressive CICATREX jackets are well supported by patients if they are made to measure, according to our procedure, but they impact the power of shoulders rotators also. The main impact of the CICATREX jacket, when worn during 4 weeks, was thus: an increase of power in shoulder rotators, mainly observed in external rotators and significant at high speed (180°/s), and an improvement in joint stability.

The VETCOSED participants were mainly women, who reported a considerable presence of joint instability, with a significant impact of the disease on daily functioning: most of them used medications (78%), among which analgesics were the most prevalent (82%), and both HAQ score ($m = 1.4$) and FIS total score ($m = 100.9$) show how important the impact is on daily life. These characteristics are quite similar to those of patients studied by Rombaut et al. [9], which illustrates that our sample was representative of this population with hEDS; and this study brings evidence for the use of compressive garments to significantly improve shoulder stability in patients with hEDS and to decrease pain, even if the decrease in pain was not significant. This downward trend in pain is nonetheless interesting, as these patients have a long history of chronic pain, and this downward trend in pain was obtained after only 1 month of wearing the jacket. This result must also be compared with the significant satisfaction reported by the participants, both for the impact of wearing the jacket on pain (78% of participants were satisfied again after 1 month) and to decrease pain, even if the decrease in pain was not significant. This downward trend in pain is nonetheless interesting, as these patients have a long history of chronic pain, and this downward trend in pain was obtained after only 1 month of wearing the jacket. This result must also be compared with the significant satisfaction reported by the participants, both for the impact of wearing the jacket on pain (78% of participants were satisfied again after 1 month) and to decrease pain, even if the decrease in pain was not significant.
patients when the procedure is not fully adapted and the garment is not comfortable.

Both external rotators and internal rotators presented weakness in our study participants at inclusion. The mean power was around 10 W in external rotators and around 20 W in internal rotators, with very big standard deviations. Regarding torque peaks, external rotators and internal rotators respectively reached around 15/27 N in our participants, whereas values in healthy participants respectively reach around 40/57 N at similar speeds [22]. This tends to underline the muscular weakness of these patients, even if it is difficult to compare them to norms, as values depend also on evaluation procedures.

The rotators are stabilizing muscles of the glenohumeral that control the proper centering of the humeral head during shoulder movements. The articular balance depends on the muscular balance and on the proprioceptive quality of the periarticular structures. By non-use or under-use, there is a loss of muscle potential around a lax joint by default of the support structures. In the context of hEDS, this deficiency of active structures is added to the deficiency of passive structures, increasing hypermobility and pain. A gain in power and strength of the external rotators improves the stability of the scapula and gives a fixed point for the use of the shoulder, improving joint stability and shoulder function. The balance of these shoulders in hEDS patients is clearly dependent on the reaction quality of the muscles.

Wearing a jacket had an impact on the power of shoulder rotators, which could be readily observed but disappeared as soon as the jacket was put off. These results are clearly in favor of

### Table 2. Immediate effect of jacket removal after 4 weeks of wear (at Day 28) on shoulder rotators kinematics, at 180°/s and 90°/s.

<table>
<thead>
<tr>
<th>Shoulder kinematics at 180°/s</th>
<th>After 4-week jacket wear period, with jacket</th>
<th>After 4-week jacket wear period, without jacket</th>
<th>Repeated measures ANOVA*</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Coefficient (SD)</td>
<td>CI 95%</td>
<td>p-Value**</td>
</tr>
<tr>
<td><strong>Shoulder kinematics at 180°/s</strong></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td><strong>External rotators power (W)</strong></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Dominant shoulder, mean (SD)</td>
<td>11.4 (9.5)</td>
<td>10.1 (8.6)</td>
<td>-1.35 (0.36)</td>
</tr>
<tr>
<td>Non-dominant shoulder, mean (SD)</td>
<td>10.8 (10.6)</td>
<td>9.3 (8.6)</td>
<td>-1.63 (0.63)</td>
</tr>
<tr>
<td><strong>Internal rotators power (W)</strong></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Dominant shoulder, mean (SD)</td>
<td>23.3 (20.9)</td>
<td>22.1 (19.4)</td>
<td>-0.37 (2.27)</td>
</tr>
<tr>
<td>Non-dominant shoulder, mean (SD)</td>
<td>22.9 (20.8)</td>
<td>20.9 (20.2)</td>
<td>-0.75 (0.33)</td>
</tr>
<tr>
<td><strong>Ratio of external/internal rotators powers</strong></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Dominant shoulder, mean (SD)</td>
<td>60 (23.7)</td>
<td>57.1 (31.5)</td>
<td>0.06 (0.1)</td>
</tr>
<tr>
<td>Non-dominant shoulder, mean (SD)</td>
<td>52.9 (24.3)</td>
<td>55 (31)</td>
<td>-0.75 (0.33)</td>
</tr>
<tr>
<td><strong>External rotators torque peaks (N m)</strong></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Dominant shoulder, mean (SD)</td>
<td>15.6 (9.3)</td>
<td>14.8 (9.2)</td>
<td>-0.66 (0.44)</td>
</tr>
<tr>
<td>Non-dominant shoulder, mean (SD)</td>
<td>15.4 (10.3)</td>
<td>14.7 (9.1)</td>
<td>-0.66 (0.44)</td>
</tr>
<tr>
<td><strong>Internal rotators torque peaks (N m)</strong></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Dominant shoulder, mean (SD)</td>
<td>27.1 (16.4)</td>
<td>26.6 (16)</td>
<td>-0.83 (0.39)</td>
</tr>
<tr>
<td>Non-dominant shoulder, mean (SD)</td>
<td>28.4 (17.1)</td>
<td>27.2 (16.4)</td>
<td>-0.83 (0.39)</td>
</tr>
<tr>
<td><strong>Ratio of external/internal rotators torque peaks</strong></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Dominant shoulder, mean (SD)</td>
<td>61.5 (17)</td>
<td>57.8 (17.7)</td>
<td>-1.42 (1.62)</td>
</tr>
<tr>
<td>Non-dominant shoulder, mean (SD)</td>
<td>55 (15)</td>
<td>55.8 (16.9)</td>
<td>-1.42 (1.62)</td>
</tr>
</tbody>
</table>

*Model adjusted on shoulder dominance and history of wearing compressive garments; **Holm–Bonferroni adjusted p-value.

SD: standard deviation; CI 95%: confidence interval at 95%.

### Table 3. Impact of 4-week jacket wear on joint stability and pain.

<table>
<thead>
<tr>
<th>Variable</th>
<th>After 4-week jacket wear</th>
<th>After 4-week without jacket</th>
<th>p-Value***</th>
</tr>
</thead>
<tbody>
<tr>
<td>Shoulder instability (decentration, subluxation or dislocation), n (%)</td>
<td>26 (72.2)</td>
<td>33 (91.7)</td>
<td>0.0163</td>
</tr>
<tr>
<td>Shoulder dislocation, n (%)</td>
<td>1 (2.8)</td>
<td>7 (19.4)</td>
<td>0.0163</td>
</tr>
<tr>
<td>VAS pain, mean (SD)</td>
<td>2.8 (2.8)</td>
<td>3.8 (2.6)</td>
<td>0.0964</td>
</tr>
<tr>
<td>Non-dominant shoulder, mean (SD)</td>
<td>2.2 (2.6)</td>
<td>3.3 (2.4)</td>
<td>0.0964</td>
</tr>
</tbody>
</table>

*Holm–Bonferroni adjusted p-values of the McNemar test for joint stability data and p-value of the repeated measures analysis of variance controlled for dominance shoulder and history of wearing garments for VAS pain.
a proprioceptive mechanism of action [13], which would promote muscle recruitment and expression of rotator power, without inducing strengthening that would persist without the jacket. Contention plays the role of articular support that deficient muscles do not allow, reassures the patient, and allows him/her to re-appropriate gestures and muscle function without risk of instability and therefore reducing pain. This conclusion should however be interpreted with caution, as VETCOSED patients only worn the compressive jacket for 4 weeks, and nobody knows until now if a longer period of use of this kind of garment could induce muscle strengthening.

The best and significant effect was found in external rotators at a high speed of isokinetic assessment (180°/s), which was an expected and positive result. Indeed, at slow speed (90°/s), the proprioceptive feedback allows the person to better control the shoulder, while at fast speed it is more complicated. But dislocations usually occur during rapid movements of daily life. So, it is interesting to observe the effect of the jacket for these fast movements. As the expected effect of the jacket was less important at 90°/s, it would have required many more patients to hope to show an effect. Lack of power is one of the limitation of this study, but EDS is an orphan disease, and the dispersion of patients over the national territory did not facilitate recruitment.

**Conclusion**

Participants reported less shoulder pain and less joint instability after having worn the compressive jacket for 4 weeks, which should be identified as great attainment for the objective of compressive jackets. In the field of hEDS, where there is a strong need for evidence-based therapy, the results of this study may support clinicians’ belief that compressive garments are effective, as long as they are worn, in promoting muscle expression of shoulder stabilizers. This finding can also be passed on to patients, who need to learn self-care skills [23] and manage the wearing of their compressive garments themselves for better effectiveness in their daily lives.

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**Disclosure statement**

No potential conflict of interest was reported by the author(s).

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