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The Language of Pain in the Hypermobile Ehlers-Danlos Syndrome: Metaphors as a Key to Understand the Experience of Pain and a Rehabilitation Tool

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The Language of Pain in the Hypermobile Ehlers-Danlos Syndrome: Metaphors as a Key to Understand the Experience of Pain and a Rehabilitation Tool

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Abstract: Ehlers–Danlos syndromes (EDS) are a heterogeneous group of heritable connective tissue disorders (HCTDs) characterized by joint hypermobility, skin hyperextensibility, and tissue fragility. Among the different types, the hypermobile EDS (hEDS) is the most frequent, and includes generalized joint hypermobility as the major diagnostic criterion. Joint hypermobility in hEDS is often associated with pain that not always allow the use of effective pain-reducing treatments. Patients with hEDS always talk about pain using a lot of descriptions. Eighty-nine patients with diagnosis of hEDS were recruited and evaluated. They were asked to write down what grief was to them. The texts were analyzed through LIWC. Correlational analyses were conducted between pain perception and language. A comparison of high/low pain perception and quality of metaphors was carried out. The results showed, depending on the level of pain perception, different language quality is evidenced. The greater the pain the lesser the positive effects and the greater the negative effects and dehumanizing metaphors. Moreover, greater pain seems to be related to a verbal experience of greater isolation and less self-care. In conclusion, the use of metaphors is a useful tool for exploring illness experience and may help clinicians in the rehabilitation program.

Keywords: chronic pain; hypermobile Ehlers–Danlos syndrome; linguistic content analysis; metaphors; pain; symbolizing process

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Introduction

Ehlers–Danlos syndromes (EDS) are a heterogeneous group of rare hereditary disorders of connective tissue (HCTDs), with common features including joint hypermobility, skin hyperextensibility, and tissue fragility [1]. The most recent clinical classification, introduced in 2017, and subsequent updates, now recognize 14 different subtypes of EDS [1–4]. The hypermobile type (hEDS) is generally considered to be the most frequent in the general population, followed by the classical (cEDS) and vascular (vEDS) types, while others appear mostly ultrarare. A detailed revision of the clinical diagnostic criteria for hEDS was also introduced in 2017, to allow a better distinction from other EDS types and other syndromic disorders that also can include generalized joint hypermobility as a major clinical feature [2]. In the meanwhile, the term "Hypermobility Spectrum Disorders (HSD)" was proposed to include clinically relevant joint hypermobility, mostly symptomatic for secondary manifestations or complications, in patients that not fulfil the clinical diagnostic criteria neither for EDS, other HCTDs, or other recognizable syndromes [5]. The diagnosis of hEDS still relies exclusively on a combination of clinical inclusion criteria (generalized joint hypermobility, chronic pain and/or joint instability, signs of multisystemic connective tissue involvement, positive family history for hEDS) and some exclusion criteria. This happens because the genetic aetiology of hEDS is still largely unknown and specific and sensible genetic tests are not currently available to support the clinical diagnosis [1]. Concerning the main symptoms of joint hypermobility in hEDS, among the others, chronic localized and/or diffuse pain is most often referred from patients and is also considered the major determinant of the perceived reduced quality of life [6,7].

Different papers have analysed chronic pain in hEDS and HSD (in the literature before 2017 the diagnosis of hEDS or Joint Hypermobility Syndrome were obtained in many patients that are expected to receive now a diagnosis of hEDS or HSD, using the current criteria and nosology) trying to explain its pathogenesis in order to better address an effective treatment [8–12]. Pain in hEDS and HSD is considered multifactorial, partly related to hypermobility, joint instability, traumas, and previous surgery, and associated with moderate to severe impairment in daily functioning [8,13]. Pain can also arise from damage to the somatosensory system itself and it should be a neuropathic pain [14]. When pain persists for more than 3–6 months or resurfaces after the tissue damage has resolved, neuroplastic changes in the pain processing pathways may lead to a hypersensitive state of the somatosensory system, a phenomenon called central sensitization [15,16].

From an observational point of view, patients with hEDS always talk about pain using a lot of descriptions and sometimes also with drawings [9]. Chronic pain is defined as pain that lasts for more than three months and is linked to many psychological comorbidities, including substance abuse, depression, and anxiety. In the absence of objective pain assessment tools, those who are experiencing it must rely on language and nonverbal pain behaviours like facial expressions to express their subjective experiences. Although grief is inherently elusive and private, often people to better succeed in communicating their internal experiences with others use metaphors, which take on symbolic significance and reflect their internal state. Allowing for external communicability and activating greater empathy of others [17]. A well-documented linguistic tool for communicating grief is metaphor [18,19]. This provides access to a more shared construction of meaning. e analysis of metaphors facilitates the exploration of how each person makes sense of the world [20].

In order to encourage the elaboration of traumatic experiences or chronic disease, the expressive writing intervention (EWI), a technique that focuses on written emotional expression, initially required participants to write about pasted-in experiences [21,22]. In later research, the writing prompts were broadened to include additional particular challenging events that had positive effects on other healthy and clinical groups [23–26]. To date there are not standard instructions for EWI but the topic should be formulated depending on the specific subjects to whom it is addressed, also with the possibility to suggest instructions focused on a present difficult experience, dedicated to positive expectations for the future, and proposing a different writing topic for each session [23]. In the present study, the writing technique was used to stimulate the production of symbolic thinking in relation to the perception of chronic pain triggered by hEDS. It was then proposed to describe the pain experience of illness through writing metaphors. Several studies have made explicit the richness that the metaphorical stimulus can induce a reorganization of affective experience and a resignification of the meaning of life [27–29].

This process promotes a different representation of the events in the memory, the memory is simplified, and it can be easily recalled to the mind, and these cognitive changes imply a different understanding of the experience and a change of perspective on the event [30]. To analyze metaphors the Linguistic Inquiry and Word Count (LIWC) [31], a computerized text analysis program, has been applied. LIWC was initially specifically created to analyze essays from EWI studies, and it is also routinely used in analyzing psychologically meaningful writings [32,33]. The LIWC program was also used to explore the association between word usage with various health and behavioral outcomes in expressive writing subjects [21]. In a recent study the LIWC was applied to written descriptions of metaphors of life experiences, finding that this process can help to reformulate own experiences [34,35]. In conclusion, LIWC represents a possibility to deeply understand its role in the writing process.

The purpose of the present work is to test how the constant perception of pain interferes in symbolic and representational processes.

Our hypothesis is that chronic pain that exceeds its tolerability produces narrative productions that are clinically different from more tolerable pain.

We have decided to analyze the use of metaphors for describe pain in patients affected by hEDS asking them to write down how they relate to their perception of their pain by responding to the following stimulus, "my pain is.....".   Standardized clinical scale has been associated.

Materials and Methods

Participants

Eighty-nine patients (79 female and 10 males, mean age 40±13,11) with diagnosis of hEDS has been recruited and evaluated. Diagnosis was made by a clinical geneticist following the 2017 diagnostic criteria and classification [2].

Measures

Linguistic Inquiry and Word Count (Pennebaker, Booth, & Francis, 2015)

The Linguistic Inquiry and Word Count (LIWC) is a computerized program aimed to analyze data related to the language use in writing reports. LIWC program includes the main text analysis module along with a group of built-in dictionaries. LIWC reads written or transcribed verbal texts, then compares each word in the text against a user-defined dictionary. After the processing module has read and accounted for all words in each text, it calculates the percentage of total words that match each of the dictionary categories. LIWC2015 v1.6 software has been used together with Italian LIWC_2007 Dictionaries. Specific word categories have been chosen for the purpose of the study: Social Processes Friends (pal, buddy, coworker) Family (mom, brother, cousin) Humans (boy, woman, group);   Affective Processes : Positive Emotions (happy, pretty, good) Negative Emotions (hate, worthless, enemy); Anxiety (nervous, afraid, tense); Anger (hate, kill, pissed) Sadness (grief, cry, sad) Past Time; Present Time; Future Time; Cognitive Processes: Inclusive (with, and, include); Exclusive (but, except, without); Personal concern: Leisure; Home; Body Care.

Clinical scale:

McGill Pain Questionnaire (MGPQ, Italian version): that measures the 'quality' and the 'amount' of pain. Only the total pain rating was considered. This value was obtained by adding the score assigned to each single pain characteristic considering the word used to represent the lowest pain intensity scored as 1, the next highest intensity as 2, and so on [36].

Zung self-rating depression scale (SDS): Zung's Self-Rating Depression Scale (SDS) has been widely used to assess depression [37]. It consists of 20 self-rated questions, each item rated on a 4-point scale ranging from 1 (a little of the time) to 4 (most of the time). The total score was acquired by multiplying the raw score by 1.25. A higher total score indicates a more severe level of depression. An SDS score of 50 (raw score = 40) suggests clinically significant symptoms [38].

Zung self-rating anxiety scale (SAS): is a scale used for discriminate anxiety from mood disorders [39]. It's a 20 item Likert scales, in which items tap psychological and physiological symptoms; each item rated on a 4-point scale ranging from 1 (none, or a little of the time) to 4 (most, or all the time). Conversion of a total scale raw score (with a potential range of 20 to 80) to a index score with a potential range of 25 to 100 is derived by dividing the sum of the values (raw scores) obtained on the 20 items by the maximum possible score of 80, converted to a decimal and multiplied by 100. Total score with a value<50 indicates a no-anxiety condition, a minimal to mild anxiety with a score between 50 to 59 points, a moderate to marked anxiety between 60 to 69 and a condition of great anxiety if the score is more than 70.

Procedure

To our knowledge, this is the first survey that investigates the use of language and metaphor in a cohort of patients affected by hEDS. Participants were recruited over a three-month period (January 2021-March 2021) through the ARCASED association (Associazione per la Ricerca, Cura ed Assistenza Sindromi di Ehlers Danlos), CEDI Onlus association (Clinici Ehlers-Danlos Italia) and AISED association (Associazione Italiana per la Sindrome di Ehlers-Danlos). All participants were included if they 1) were 15 years of age or older, 2) had a diagnosis of hEDS, 3) spoke Italian language.

The survey was custom-built and self-administered taking about fifteen minutes to be completed. It was made clear to participants that it was anonymous and voluntary. The survey was hosted on Google Form which automatically collected the answers in order of arrival and transferred them onto a spreadsheet.

The final version of the questionnaire addressed the following areas: (a) demographic questions including age, identifying gender and current employment status, (b) pain data collection through the McGill Pain Questionnaire, (c) metaphors used to define pain - in order to minimize the conceptual ambiguity, the general definition of metaphor was clarified and two examples were provided – and (d) psychopathological correlates through Zung Self-Rating Anxiety Scale (SAS) and Zung Self-Rating Depression Scale (SDS).

Statistical Analyses

All statistical analyses were performed using the Statistical Package for Social Science version 24 (SPSS version 24, Armonk, NY). Data are reported as means and standard deviation for continuous variables and as percentage for discrete variables. To analyze metaphors in relation to pain experience, the group was divided using the cut-off of 35 points for the MGPQ total index that is the mean value of pain in a population of patients with primary fibromyalgia [40], in order to differentiate into two group: The first with high pain (MGPQ≥36) and the second with low pain (MGPQ≤35).

Independent T test has been applied to compare linguistic measures. Pearson correlation has been used to verify relation between linguistic measure and symptoms measures. A p value < .05 was considered significant.

Results

Eighty-nine patients were evaluated; the mean age was 40 (min 15 max 71) with a prevalence of females (79 F/10M).

The mean value of the different scale used to evaluate pain, anxiety and depression of these patients are summarized in Table 1.

Table 1. The mean value of the MGPQ, The Zung Anxiety scale and the Zung depression scale.

MGPQ	**Total index (mean ±s.d.)**	N° patients with High pain (≥36) /%		N° patients with Low pain (≤35) /%	
	46,65 ± 13,55	71/79,8		18/20,2	
	Mean value	N° patients <50 (absent)/total %	N° patients 50-59 (minimal to mild)/total %	N° patients 60-69 (moderate to marked)/total %	N° patients >70 (great)/total %
Zung Anxiety	64,23 ±11,73	10/89 11,24%	19/89 21,34	30/89 33,71	30/89 33,71

score					
Zung Depression score	61,03 ± 12,09	18/89 20,21%	20/89 22,5%	26/89 29,2%	25/89 28,09%

Using the pain cut-off indicated, we divided the sample into two groups and analyzed LIWC language measures. The hypothesis developed was that chronicity of pain perception afflicts patients by conditioning the way they develop symbolic and linguistic processes. This hypothesis of ours can be said to be confirmed. The use of metaphors in the clinical group for pain perception turns out to be significantly different from the group with low pain perception for many linguistic parameters (Table 2).

Table 2. Independent T test in linguistic measures of LIWC indexes, between "Low-High pain perception".

Variable	**High Pain (n.71)**		**Low Pain (n.18)**		**T**	**P**
	M	**ds**	**M**	**ds**		
LIWC						
Friends	0,28	1,41	2,17	4,51	-3,024	**.003***
Family	0,02	0,17	0,00	0,00	0,501	.617
Human	0,24	1,01	2,42	4,78	-3,601	**.001***
Positive Emotions	0,00	0,00	0,70	2,02	-2,945	**.004***
Negative Emotion	3,90	6,64	2,63	5,16	0,758	.451
Anxiety	0,16	1,04	0,23	0,98	-0,260	.796
Anger	1,03	2,51	0,25	1,07	1,288	.05
Sadness	1,89	4,16	0,00	0,00	-2,645	**.006***
Past Time	0,56	2,58	0,15	0,66	0,660	0,511
Present Time	4,76	5,83	4,28	5,96	0,314	0,755
Future Time	0,00	0,00	0,21	0,87	-2,021	**.007***
Inclusive	0,72	1,64	0,00	0,00	3,684	**.000***
Exclusive	6,39	6,48	4,33	5,720	1,234	.221
Leisure	0,26	1,33	1,40	4,29	-1,908	**.036***
Home	0,13	1,08	1,39	4,28	-2,237	**.028***
Work	0,210	0,70	0,89	2,07	-2,182	**.032***
Body care	0,000	0,00	0,666	1,020	-1,974	**.033***
Greed of freedom 87; *p.< .05						

Bivariate correlation analysis on the whole sample N. 89 between the linguistic and clinical measures of anxiety, depression and pain scale specifically show that the depression scale correlates negatively with positive emotions (r -.252 p.< .05) and body-care (r -.243 p.<.05) while positively with anger (r 243 p<.05). The anxiety scale correlates negatively with friends (r -233 p<.05), human (r -305 p<.05) and body-care (- 256 p<.05). Finally, pain correlates negatively like anxiety with the same language categories, friends (r -245 p<.05), human (r -305 p<.02) and body-care (r- 441 p<.01).

Discussion

All patients were asked to respond in writing to the following question trying to make explicit a metaphor indicating " My pain is...". Correlation analysis shows that the clinical scales of anxiety, depression and pain are related to specific scales of the human/non-human relationship. Depression is related to anger and lower positive feelings. Element consistent with depressive dimension [41]. Anxiety and pain perception correlate with the same scales highlighting how greater anxiety and greater pain tend to bring out a more alien and dehumanized narrative. Deepening this data by comparing the two groups this element is confirmed. The groups with high and low pain perception used this stimulus in a significantly different way. The lower perception of pain is expressed by the metaphor of a presence in life as a friend/partner to live with. It is identified as a "companion" a friend who never leaves you but to have been tolerate. This consistently repeating description is what leads to differentiating the low pain perception group with more words of Human, Friend, positive emotions. “*A burning fire…A dull and annoying travel friend*” or “*My partner…sometimes silent sometimes not*” or “A troublesome companion in life: “*When it is most acute, I need to take care of him in my daily life…A troublesome master of life*”. In other words, a more tolerable perception of pain leads it to be experienced as a friend who is loved and cared for. This element is confirmed both in the correlations and in the difference between groups. Once pain exceeds the tolerability indices, however, the situation changes completely. Pain is metaphorized as a stranger, a dehumanizing element, preventing pleasure, activities. Finally, the *inclusive* category denotes the impossibility of excluding it from one's life. Pain is in fact an element that connotes existence. In patients with clinical pain perception, they represent pain as an element to be kept "out" to fight, an intrusive presence: “*A rodent eating viscera*”, “*A weight crushing me, a corset/armor constricting from head to toe*” or again “*Ice on my face*". Thus, it seems that the level of chronicity of pain perception in the metaphorical dimension drives an inside/outside struggle, where pain crosses the tolerance level it becomes an invader. The prevailing linguistic metaphor of the group with more pain is a dehumanized experience, pain from a life companion becomes the enemy from which to succeed in freeing oneself. An enemy so strong that it ruins one's life and one's pleasure. It could be concluded that the pain metaphors in this group of patients with a chronic illness represent how the relationship with pain as a love affair, that is, it can be tolerated with flaws, but past a certain limit it becomes the enemy to be divorced from or to died (Table 3).

Table 3. Examples of metaphors for the low pain and high pain groups “My pain is…”.

LOW PAIN GROUP	HIGH PAIN GROUP
an annoying companion of life	a bomb bursting inside
mine, like a brand. suppressible with medicine, tolerated and listened to now that i know why. i see it change, listen to it move, like an ant	a knife that haunts
light but pressing in time after a breathless intense bump	an acquaintance met by chance
now a part of me, of my life, every single day, light or heartbreaking i know it will be there like the sun rising in the sky every morning	like waiting for the time to come to live and die
my companion, sometimes silent sometimes not	my pain is like a knife that pierces me
swinging in frequency a companion on the journey	my pain is like the end of the world

a fire that burns a traveling companion dull and annoying	i feel fragile like glass as if i have to be always careful of every movement, and every crack of my joints
POWERFUL AS A FIRE	like having machines running over you
a harassing master of life	the feeling that my joints are gears destined to come apart at any moment

Conclusions

Our study confirmed that patients affected by symptomatic (generalized) joint hypermobility, with clinical diagnoses of hEDS, try to use metaphors to better succeed in communicating their pain experience. Pain is described with strong metaphors and the more dehumanizing felt the stronger and more chronic the pain experience. Thus, a close relationship seems to emerge between pain perception and the quality of metaphor content. This finding agrees with previous studies in which the use of symbolic language allows for embodied communication and a greater emotional vehicle to the other [42–44]. Language per se is duenque an element that allows for a possible reframing with the experience of illness, with the communicability of one's pain.

Language therefore is a therapeutic vehicle, which could be used for interventions aimed at integrating the mind-body relationship.

In this study, patients with a higher level of pain tended to use emotional language with external references ("a knife," "a bomb") that might reflect a sense of helplessness and detachment from their bodies, as they had to resort to describing it as an aggression from the outside rather than an inner discomfort. These findings are consistent with previous research on pain metaphors [45,46], and it is likely that the personification of pain as an external force gives patients the opportunity to create an objectifiable enemy to fight, thus isolating the sick part from the healthy part within the body.

In contrast to Munday's findings [46], none of our participants referred to everyday experiences of pain (such as illness or accidents), as if to argue that common events shared by listeners could not adequately describe the complexity of chronic pain.

Integrated interventions aimed at the expressiveness of the internal world and symbolization of disease processes would allow for transformative thinking and experiencing of pain. This study highlights how refining psychological interventions aimed at storytelling, and expression of one's pain experience would facilitate sharing and adherence to treatment [47,48]. The findings confirm that chronic pain has a complex and disorganizing impact on people's lives making also difficult to organize a rehabilitative treatment–

In rehabilitation field, the use of metaphors is often used but not always interpreted; if used is instead an important tool that may help rehabilitator to understand the type of pain and especially organize the exercise program; pain has a strong correlation with cognitive, sensitive, emotional elements and of memory, often is the only element for the patient to recognize the own body, so exercise proposed to the patients, needs to have cognitive elements [49], able also to reduce fatigue and improve finction [51].

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