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They’ve been BITTEN: reports of institutional and provider betrayal and links with Ehlers-Danlos Syndrome patients’ current symptoms, unmet needs and healthcare expectations

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Abstract

Introduction: Patients with rare and/or care-intensive conditions, such as Ehlers-Danlos Syndrome (EDS), can pose challenges to their healthcare providers (HCPs). The current study used the BITTEN framework[1] to code EDS patients’ open-ended written responses to a needs survey to determine their self-reported prevalence of healthcare institutional betrayal and its link with their expressed symptoms, provider perceptions, unmet needs, and on-going healthcare-related expectations.

Methods: Patients with EDS ($n = 234$) were recruited *via* a rare disease electronic mailing list and snowball sampling. A total of one-hundred and six respondents (45.3%) endorsed having unmet healthcare-related needs; of these, 104 (99%) completed an open-ended prompt about these needs. Responses were coded for components of BITTEN, a framework designed to link patients’ past, current, and future healthcare-related experiences in a trauma informed manner.

Results: Many respondents with ongoing needs endorsed experiencing past institutional and provider betrayal (43%; $n = 45$), current mental health symptoms (91.4%; $n = 95$), negative expectations for future healthcare (40.4%; $n = 62$), and a lack of trust in their healthcare provider (22.1%; $n = 23$). There were no significant differences in post-traumatic stress disorder (PTSD)/anxiety, depression/sadness, or isolative symptoms between respondents coded for institutional betrayal ($n = 45$) compared with those not ($n = 59$). However, EDS respondents reporting institutional betrayal were significantly more likely to self-report anger and irritability symptoms, a lack of trust in their HCPs, and more negative expectations for future healthcare than those not reporting institutional betrayal.

Discussion/conclusions: The frequent spontaneous reporting of past healthcare betrayals among patients with EDS implies the need for trauma-informed care and provider education. Given that experiences of institutional betrayal are associated with increased anger and irritability, as well as with negative expectations for future healthcare interactions, efforts to repair healthcare provider and system-wide relationship ruptures might have positive healthcare consequences.

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Plain language summary

Reports of Institutional and Provider Betrayal and Links with Ehlers-Danlos Syndrome Patients' Current Symptoms, Unmet Needs and Future Healthcare Expectations

What is EDS?

Ehlers-Danlos Syndrome (EDS) refers to a group of rare genetic connective tissue disorders that are primarily characterized by skin hyperelasticity, joint hypermobility, and tissue fragility. Connective tissue is largely responsible for the structural integrity of our bodies, and there are several EDS subtypes which each describe a specific connective tissue problem. In addition, there is significant overlap between EDS types and other kinds of connective tissue disorders. As a result, recognizing, diagnosing, and treating EDS is often challenging.

What is Institutional betrayal?

Institutional Betrayal here refers to a harmful action (i.e. commission) or lack of action (i.e. omission) on the part of a healthcare institution, individual provider/healthcare team, or insurance company. When a patient trusts that the healthcare system will act in their best interest, and trust is violated, institutional betrayal occurs.

What is BITTEN?

BITTEN is an acronym for Betrayal, Indicator, Trauma symptoms, Trust, Expectations, and Needs. It is a framework meant to capture previous problematic healthcare-related experiences in EDS patients, then to consider how those experiences influence a patient's current symptoms, provider trust, future expectations in healthcare encounters, and on-going needs.

Why was this done?

EDS, like many rare diseases, is hard to recognize and manage. We aim to:

- Give voice to EDS patients and their common unmet needs and healthcare-related expectations.
- Highlight how healthcare providers can apply BITTEN to improve care practices in rare disease patient encounters.

What did we do?

Using a newly articulated applied model of healthcare, BITTEN, we analyzed the open-ended responses of EDS patients describing their unmet emotional and mental health needs.

What did we find?

- Nearly half of EDS patients who indicated they had unmet needs reported experiencing institutional betrayal.
- EDS patients who reported institutional betrayal also expressed anger, a lack of trust in healthcare providers, negative expectations for future healthcare, and more unmet needs more frequently than EDS patients who did not report institutional betrayal.

What does this mean?

- The EDS patients in this sample were not directly asked if they had experienced institutional betrayal, so the exact prevalence is not known. Furthermore, responses were obtained voluntarily *via* the internet, so caution should be taken when generalizing these findings.
- However, results indicate that too many patients with EDS have experienced healthcare betrayals; these experiences are associated with current anger and negative expectations for future healthcare interactions.

- The prevalence of past negative healthcare experiences, along with current unmet needs and future negative healthcare expectations in EDS patients who have experienced institutional betrayal, highlights the need for healthcare providers to tend to these experiences, mend patient-provider barriers, and provide higher quality healthcare.

Keywords: EDS, Ehlers-Danlos Syndrome, healthcare, informed trust, institutional betrayal, nursing theory, patient centered care

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Introduction

Healthcare workers manage the acute, chronic, and preventive healthcare needs of a myriad of patients, many presenting with sociodemographic, diagnostic, and physical health complexities.[2] These healthcare providers (HCPs) often face the challenge of providing comprehensive care to patients with complex needs and coordinating care among specialty practitioners, while also managing a fast-paced acute care practice. Though inherently rewarding, providing ongoing care can be difficult, particularly to patient populations who suffer with a relatively rare disease that is not well known to and often not recognized by many HCPs. Patients suffering from Ehlers-Danlos Syndrome (EDS), a group of painful connective tissue disorders, often present such complexities. EDS is difficult to diagnose; it requires a complex, multi-pronged, and sustainable treatment plan; in addition, it lacks a designated specialty provider. EDS patients may then struggle with seeking and obtaining disease-attuned care when being treated in a variety of community and clinical healthcare settings.

EDS has thirteen subtypes (e.g. classical, hypermobile, vascular). Each subtype has a diverse clinical presentation and most require additional testing to confirm their respective genetic abnormality.[3] In addition, EDS symptoms often overlap with other connective tissue disorders, making disease comorbidity and diagnostic uncertainty prevalent.[3] As a result, misdiagnosis and diagnostic frustration are common experiences of EDS patients and their HCPs.[4] These experiences may influence EDS patients' current healthcare-seeking behaviors and patient-provider trust in ways that need to be identified and addressed.[5] However, HCPs have lacked a comprehensive framework to concisely organize and respond to the physical and mental health needs and expectations of patients with complex chronic conditions or rare disease presentations, such as EDS.[5]

To this end, this study aims to utilize a user-friendly, applied, trauma-informed framework, BITTEN,[1] to identify, code, and understand the lived-experiences of EDS patients. Specifically, BITTEN1 was employed to document EDS patients' past problematic healthcare experiences (especially experiences of institutional betrayal), and to consider how institutional betrayal may relate to patients' ongoing unmet mental health symptoms, needs, and healthcare-related expectations. Healthcare-related institutional betrayal is defined as the enactment of a harmful action (i.e. commission of a betrayal behavior) or failure to engage in an expected protective action (i.e. omission of care behavior) on the part of an individual provider/healthcare team, healthcare institution, or insurance company. To be considered institutional betrayal, the behavior(s) must be experienced as a violation of the patient's presumed trust that the healthcare provider/team or healthcare system will act in the patient's best interest.[6]

According to the BITTEN framework, the patient arrives at the healthcare appointment with a symptom indicator or index event, which traditionally forms the primary (if not exclusive) motivation for seeking healthcare. However, using BITTEN, the HCP should first consider the likelihood that this indicator will trigger the patient's potential trauma history (e.g. a urinary tract infection may be acute and straightforward, but EDS often entails a complex, sensitive medical history). After weighing the relevance of the indicator, then noting any current presence of emotional or trauma symptoms (anxiety, sadness, irritability), the HCP should consider whether institutional betrayal (IB) and/or relevant traumatic experiences may have occurred prior to the encounter. It should be noted that, while IB is a specific type of trauma or betrayal, traumatic experiences may also include medical trauma and non-healthcare-related traumatic events (e.g. sexual assault, adverse childhood experiences).[7] According to BITTEN, both IB and other traumatic experiences may then generate trauma symptoms and influence a patient's trust in their healthcare team;[1] the two 'Ts' of BITTEN. A negative cascade of trauma symptoms and trust ruptures are most likely to manifest in response to a related indicator or when seeking care within the institution in which the IB took place.[1] Theoretically, these two 'Ts' of BITTEN will, in turn, influence the patient's expectations for future healthcare encounters and their current and future healthcare needs.

In the context of EDS patients, the BITTEN framework postulates that not only may an EDS patient's current healthcare encounter experience influence future healthcare encounters, but that undetected past trauma experiences and IB may also influence current encounters. BITTEN posits that traditional 'indicator only' healthcare encounters may fail to account for EDS patients' historical IBs, traumas, and trauma symptoms, all of which have direct bearing on their current HCP trust levels, care-related expectations, and unmet health-related needs. This supposition is tested in the current study. Specifically, qualitative, open-ended written responses were collected from EDS patients regarding their unmet mental health needs to: a) give voice to the experiences of EDS patients and their common unmet needs and healthcare-related expectations, and b) highlight how healthcare providers can apply BITTEN to improve care practices in rare disease patient encounters. For these purposes, patient responses were coded according to the BITTEN framework (e.g. the presence of institutional betrayal [omission/commission]; indicator, experience of trauma and trauma symptoms; trust in provider; future expectations for healthcare; and unmet needs).

Five aims were addressed to determine:

1. The prevalence of institutional betrayal in EDS patients' open-ended responses.
2. The prevalence of trauma (medical), trauma-related and/or emotional symptoms [anxiety/post-traumatic stress disorder (PTSD), depression/sadness, anger/irritability, isolation/loneliness], and HCP trust ruptures.
3. EDS patients' self-reported expectations (provider incompetence, gaslighting or psychiatrization,[8] adverse medical outcome, self-advocacy, belief that nothing will change) for future care.
4. EDS patients' unmet healthcare needs (physical health needs, mental health needs, HCP education, and social services).
5. Per the BITTEN framework, whether there are increased self-reports of medical trauma, trauma symptoms, HCP trust ruptures, or different expectations and needs between EDS patients with and without self-reported institutional betrayal.

Method

Participants and Procedure: Purposive sampling was used to recruit EDS participants *via* the Ehlers-Danlos Society International Listserv, which is connected with this website (www.ehlers-danlos.com).[9] Contact with the Ehlers-Danlos Society International was initiated by the second author who is a registered nurse with a PhD. No prior relationship existed with the Ehlers-Danlos Society International. Snowball sampling then occurred *via* forwarded ListServ emails. Study information was posted to the ListServ along with an online Qualtrics survey link. The survey's Qualtrics link was visited 317 separate times. After clicking the link, respondents read information about the study and learned of the study's age requirement (i.e. 19 years of age or older). After reading an Institutional Review Board (IRB)-approved study information sheet, participants had to electronically agree to continue with the study before being given access to the survey. The first 45 individuals who met study criteria, participated, and provided a valid U.S. address, were told they would receive a $25 gift card. Inclusion criteria for the study were: 1) age 19 years or older; 2) officially diagnosed with EDS; 3) able to read and write in English; 4) US resident receiving US healthcare. Failure to qualify on any of these four criteria resulted in study exclusion ($n=25$), with one participant having a reported age less than 19 ($n=1$); and the remainder living internationally or receiving international healthcare ($n=24$). Addresses were collected *via* a separate, unlinked survey. As per approved IRB protocol (expedited IRB approval #1235568), consent was obtained electronically; participation was voluntary and anonymous. Participants could choose not to answer any question. Post-data collection, 13 more surveys were excluded due to extensive missing data.

As a result, the final sample consisted of 234 participants who self-reported as having EDS and who were receiving healthcare in the United States. The majority (94%) of participants were female ($n=220$). The mean age of respondents was 39.1 years old, with an age range of 19–70. The average age at diagnosis of EDS was 33.1 years. The majority (77.6%, $n=180$) of patients indicated they were diagnosed with hypermobile EDS. Of these 234, 45.2% ($n=106$) endorsed having unmet mental health needs on a binary item. Of those, 104 participants (99%) provided written qualitative responses to a follow-up open-ended prompt: "You have indicated that you have emotional or mental needs that you feel are not well managed. Please describe your emotional or mental needs that are not well managed." These qualitative responses are the focus of the current mixed methods study; the population of provided responses were coded for this project.

Materials and methods

Survey: The open-ended prompt was embedded within a larger online survey that consisted of 39 items assessing patients' needs based on the eight domains of palliative care (e.g. physical, social, psychological, ethical/legal).[10]

Coding Process: We used qualitative content analysis to consider the relationships between reports of institutional betrayal and trauma symptoms, trust reduction, healthcare expectations, and ongoing needs as expressed in the written responses to the one open-ended question. In directed qualitative content analysis, codes are derived from extant theory.[11] Informed by the BITTEN framework, pre-existing domains served as initial coding categories. In order to create theoretically-based definitions of categories for codes, a BITTEN coding manual was developed to score participants' qualitative responses. Two postgraduate research assistants (RAs) were then trained in the coding scheme. Prior to coding, the RAs familiarized themselves with the data by reading participants' responses. Next, the RAs piloted the coding scheme in an initial batch of responses ($n=25$). Initial coder agreement was calculated and determined to be excellent (93%). Any divergent codes were reviewed in conjunction with the first author to obtain consensus. This deductive process was repeated sequentially, with a final overall agreement of 94%. In addition to identifying codes according to the BITTEN model, the authors were interested in identifying the prevalence of betrayal in EDS patients, and accordingly, instances were identified. These are presented in Table 1. Member checks were not conducted, as participants' responses were provided anonymously and via written electronic communication (Qualtrics). Consensus codes are presented (Table 1).

Results

Statistical analyses: Analyses were conducted using SPSS Version 26. To address aims 1 and 2, frequency analyses were conducted to describe the prevalence of institutional betrayal, medical trauma, particular trauma symptoms, and trust ruptures with HCPs appearing in the spontaneous responses of EDS patients. To address aim 3, relevant data were coded to identify healthcare expectations. Five types of healthcare expectations emerged from the data and were subsequently defined. During coding, a sixth category (other") was needed to capture expectations that were not easily coded into one of the five existing types of expectations. Similarly, to address aim 4, four discrete and commonly-occurring needs were expressed by EDS patients. Coders utilized these four options and were provided an additional 5th "other" category for less commonly occurring needs. Data saturation was determined to have been obtained.[12] Finally, aim 5 considered differences in responses between patients who were coded as reporting IB *versus* those who did not indicate IB in their open-ended responses. To make these determinations, a series of Chi–Square analyses were conducted. Given the exploratory nature of these analyses, a p-value $\leqslant 0.05$ was retained to determine significance.

Indicator: Qualitative responses were coded for indicator(s) (e.g. EDS, pain, mental health,

Table 1. The prevalence of institutional betrayal, trauma exposure, mental health symptoms, unmet needs, and future healthcare expectations coded within the written responses.

Experience self-reported	% EDS patients with experience	*n*	Aggregate example phrase
Institutional betrayal experiences	48.0	50	"Nobody I've seen even knew what EDS was" (*omission*); "My pain is always blamed on mental causes" (*commission*)
Isolated commission	6.7	7	
Isolated omission	11.5	12	
Systemic commission	17.3	18	
Systemic omission	23.1	24	
Insurance difficulties	6.7	7	
Indicator/index event	99.0	103	"My EDS symptoms alongside my other health issues are becoming increasingly debilitating in everyday life." (*EDS, other*)
EDS	33.7	35	
Pain	20.2	21	
Mental health	83.7	87	
Addiction	1.0	1	
Other	18.3	19	
Trauma exposure	7.7	8	"The sheer number of complications due to this last misdiagnosis have left me anxious and reluctant in seeking future healthcare...but I'm still scared to face this alone." (*Medical trauma, anxiety, loneliness*)
Medical trauma	5.8	6	
Interpersonal trauma	1.9	2	
Trauma and mental health symptoms	91.4	95	
Anxiety, stress, PTSD, fear	59.6	62	
Depression, sadness, hopelessness	54.8	57	
Tiredness, exhaustion	8.7	9	
Anger, irritability, annoyance	15.4	16	
Loneliness, isolation	17.3	18	
Trust in HCP	26.9	28	"My doctor is doing their best, but my psychiatrist is less understanding." (*Negative, positive trust*)
Negative	22.1	23	
Positive	6.7	7	

(Continued)

Table 1. (Continued)

Experience self-reported	% EDS patients with experience	*n*	Aggregate example phrase
Expectations	40.4	62	"Most visits it feels like I need to fight to be heard. . . Otherwise they don't seem to care enough." (*Self-advocacy, symptoms dismissed*)
Provider incompetence	20.2	21	
Not being believed/symptoms dismissed	19.2	20	
Poor care/medical harm	10.6	11	
Self-advocacy	22.1	23	
Nothing is going to change	24.0	25	
Needs unmet	99.0	103	"I wish my doctors better understood the pain that comes with my subtype. It can be hard to bear." (*HCP education, physical health*)
HCP education/self-education/insurance changes	30.8	32	
Physical health	51.9	54	
Mental health	87.5	91	
Social needs	22.1	23	

EDS, Ehlers-Danlos Syndrome; HCP, healthcare provider; *n*, number of patients; PTSD, post-traumatic stress disorder.

addiction, other) associated with participants' unmet emotional or mental health needs and future healthcare expectations. In almost all cases (99%), it was possible to code for an indicator (see Table 1). It should be noted that, because many participants reported more than one reason for seeking healthcare, category response totals may exceed 100%. As expected, given the prompt, the most common indicator reported in conjunction with an unmet need was a mental health concern ($n=87$; 83%). Other common indicators were EDS symptoms ($n=35$; 34%) or pain ($n=21$; 20%).

Institutional betrayal: In accordance with BITTEN,[1] responses were coded for mention of IB. Almost half reported an institutional betrayal experience ($n=45$; 43%) with the most common being systemic omission ($n=24$; 23%). Examples of systemic omission include a healthcare system-wide lack of knowledge about their specific disease, or when their HCP or team failed to proactively educate themselves about their patients' disease. EDS participants also reported occurrences of IB systemic commission ($n=18$; 17%). 'Gaslighting', an IB commission, was the most commonly reported IB. EDS participants described 'gaslighting' as being told that their physical EDS symptoms were not medically possible or were best explained by mental health concerns, a process also known as psychiatrization,[8] in the absence of knowledge about EDS.

Trauma: Few respondents ($n=8$) spontaneously reported additional traumatic events. When mentioned, however, medical traumas ($n=6$; 6%) were three times more likely to be reported than interpersonal traumas ($n=2$; 2%).

Trauma symptoms: While specific traumas were rarely mentioned, more than 90% of EDS respondents' written comments ($n=95$) contained information that could be coded as a PTSD symptom or a related mental health concern. Specifically, 60% of responses referred to ongoing symptoms of stress, anxiety, PTSD, and/

or fear ($n = 62$). Likewise, over half of responses ($n = 57$; 55%) referenced feeling depressed, sad, or hopeless in relation to their EDS healthcare. Increased anger, annoyance, and/or irritability were self-reported by 15% of respondents ($n = 16$). Again, respondents could self-report more than one symptom, so percentages may total greater than 100%.

Trust in provider: Over three times as many respondents indicated a lack of trust in their current HCP or healthcare team (e.g. a trust-rupture) as compared with those highlighting positive trust (22% *versus* 7%).

Expectations for future healthcare: Forty percent of respondents' open-ended responses could be coded for expectations for future healthcare encounters; future expectations were almost universally negative. One in every four respondents ($n = 25$) expressed some degree of hopelessness regarding future encounters. Similarly, 22% ($n = 23$) believed that they would 'have to advocate' for themselves to receive reasonable care. Roughly 20% of respondents ($n = 21$) expected to encounter provider incompetence regarding their EDS treatment, and 19% ($n = 20$) expected their future symptoms would be ignored, dismissed as unimportant, or disbelieved. Furthermore, 11% of respondents ($n = 11$) overtly worried that future healthcare encounters would result in poor care, medical errors, or physical harm.

Needs: As expected based on the prompt, 99% of responses ($n = 103$) described having unmet healthcare-related needs. The majority (88%) noted unmet mental health needs ($n = 91$), while 52% reported unmet physical health needs ($n = 54$). Furthermore, 31% of responses ($n = 32$) indicated a need for their provider and/or insurance company to be better educated about EDS. In addition, 22% of responses ($n = 23$) indicated that the nature of EDS generated unmet social support needs. While specific social support needs varied, respondents frequently cited experiencing loneliness and isolation, and needing more support from family and friends.

Lastly, as theorized by the BITTEN theoretical framework and depicted in Table 2, respondents who reported IB ($n = 45$) were compared with those whose responses did not contain IB ($n = 59$). Those coded for IB were significantly more likely to also report a medical trauma *versus* those not coded for IB [(13.3% *versus* 0%), Chi–Square (1) = 8.35, $p = 0.004$]. Although there were no significant differences between the two groups in reports of PTSD/anxiety, depression/sadness, or isolative symptoms, the group reporting IB was significantly more likely to report anger and irritability [(24.4% *versus* 8.5%), Chi–Square (1) = 5.00, $p < 0.05$]. There were also group differences in lack of trust in their HCP [(51% of the EDS group with IB *versus* 0% of the no-IB EDS group), Chi–Square (1) = 38.71, $p < 0.001$].

Also consistent with the BITTEN model, four of the five expectations for future healthcare encounters were elevated in the EDS group with IB, compared with the no-IB EDS group. EDS respondents with IB were significantly more likely to expect provider incompetence (47% *versus* 0%) and believe that they would receive poor care or experience medical harm (22% *versus* 2%). EDS respondents with IB were also significantly more likely to expect that their provider would not believe them (i.e. 'gaslighting' or psychiatrization) (38% *versus* 5%) and that they would need to self-advocate (40% *versus* 9%), all p's < 0.05. However, both groups were equally likely to express the expectation that 'nothing will change' and they will just 'have to deal' with their disease (24% *versus* 19%). Similarly, on-going physical and mental health care needs did not significantly differ between groups. However, the EDS group with IB was more likely to express a desire for their providers, the healthcare system, and insurers to be more educated about EDS [(31% *versus* 9%), Chi–Square = 31.8, $p < 0.001$].

Discussion

Current findings were generated *via* coding the written qualitative responses of EDS patients to an open-ended prompt about their unmet mental health needs. Nearly all participants who indicated having unmet mental health needs answered in detail, creating a rich resource of EDS patients' lived healthcare-related experiences. Moreover, although the survey did not explicitly inquire about patients' experiences related to the BITTEN theoretical framework,[1] the overwhelming majority of respondents spontaneously reported experiences captured by BITTEN. Furthermore, many of the predicted relationships among components of the BITTEN model were obtained, suggesting the model's promise for helping HCPs better understand this rare disease patient population.

Table 2. The relationships between institutional betrayal and trauma/emotional symptoms, trust in provider, expectations, and needs as spontaneously self-reported by EDS patients.

Overall category	Code	Institutional betrayal coded *n* = 45 (%)	No institutional betrayal *n* = 59 (%)	Chi–square value df = 1	*p*-value
Co-occurring trauma	Medical trauma	13.3	0	8.35	0.004
Trauma/Emo symptom	Anxiety, stress, PTSD	64.4	55.9	<1	0.38
	Depression, sadness, hopelessness	55.6	54.2	<1	0.89
	Anger, irritability	24.4	8.5	5.00	0.02
	Isolation	13.3	20.3	<1	0.35
Provider trust	Lack of trust	51	0	38.72	0.000
Expectations	Provider incompetence	46.7	0	34.50	0.000
	Not being believed, gaslighting of symptoms	37.8	5.1	17.57	0.000
	Poor care/medical harm/ adverse outcome	22.2	1.7	11.37	0.001
	Pt will need to self-advocate	40.0	8.5	14.73	0.000
	Nothing will change/hopeless/ deal with it	24.0	18.6	2.17	0.14
Ongoing needs	Physical health	62.2	44.1	3.37	0.07
	Mental health	86.7	88.1	<1	0.82
	Education and coverage	30.8	8.5	31.82	0.000

n, number of patients; PTSD, post-traumatic stress disorder.

Specifically, institutional betrayal was quite prevalent among EDS participants, with roughly half of respondents to the prompt endorsing IB. Reported IBs included HCPs' failure to acquire adequate knowledge of EDS itself, or providers' insistence that patients' symptoms had psychogenic rather than EDS-related causes. Patients commonly described being 'gaslighted' by their providers in ways that reduced their overall quality of care and were associated with patient-provider trust ruptures. The process of trying to present one's symptoms as credible to reduce psychiatrization is more likely to happen to women seeking medical care.[13] It should be noted that the majority of EDS patients are women; therefore, they are disproportionately at risk.[14]

Although few EDS participants spontaneously mentioned a specific trauma, trauma-related mental health symptoms (e.g. anxiety/PTSD, depression/sadness, anger/irritability, isolation/loneliness) were referenced in over 90% of responses. It is important to note that although these mental health symptoms may result from trauma exposure, co-existing physical problems, mental health conditions, and life stressors may also contribute to symptom presence. Future longitudinal studies are needed to disentangle the relations among mental health symptoms, EDS, trauma exposure, and negative healthcare experiences.

Despite not being asked about their relationship with their current HCP(s), EDS respondents frequently noted specific trust ruptures, as well as a general distrust in their provider(s). This supports the notion that EDS patients are more prone to trust difficulties with their providers than the general population;[15] perhaps due to the medical community's general lack of knowledge about EDS, and hence, EDS patients' notoriously tumultuous diagnostic journey and complex treatment processes. There were also group differences in lack of trust in their HCP, with approximately half of EDS respondents with IB reporting this compared with none of the no-IB EDS respondents. Future studies should examine whether the general lack of provider trust coded in the current study occurs in other patient populations with rare, chronic diseases (e.g. myasthenia gravis, cyclic vomiting syndrome) or is specific to EDS patients. In addition, because respondents' sociodemographic data were not collected in the current study, further work should consider how both vulnerable and marginalized patient populations experience HCP trust in the context of systemic racism, sexism, and other prejudicial attitudes and behaviors that may be manifesting across society and within healthcare practice.

As predicted by the BITTEN theoretical framework, EDS respondents' past experiences were associated with their future expectations and healthcare needs. It should be noted that EDS respondents with IB were significantly more likely to both recall and anticipate negative healthcare experiences. For example, EDS respondents with IB were more likely to mention a medical trauma compared with EDS respondents not coded for IB. While there were no significant differences in expressed PTSD/anxiety, depression/sadness, or isolative symptoms, which were generally prevalent, the EDS group reporting IB was significantly more likely to report anger/irritability compared with the no-IB EDS group. This is noteworthy, as this emotional response might make positive supportive provider-patient relationships more difficult.

Also consistent with BITTEN, four of the five negative expectations for future healthcare were elevated in the EDS group reporting IB. Specifically, EDS respondents with IB were significantly more likely to expect provider incompetence, future receipt of poor care/medical harm, and to be gaslit or disbelieved by their HCP. They also expected to have to self-advocate. Furthermore, EDS respondents with IB were also more likely than their no-IB peers to express a desire for providers, the overall system, and insurers to be better educated about EDS.

However, regardless of IB history, EDS respondents were equally likely to expect that nothing will change, and they will just 'have to deal' with their disease. Likewise, EDS respondents with and without IB histories self-reported similar physical and mental healthcare needs. These findings highlight the challenges that all patients with this rare disease are likely to face as they navigate a healthcare system that does not yet have clear and effective treatment guidelines for their hard to diagnose and hard to manage disease process.[5]

Several study limitations must be noted. Respondents were recruited *via* a listserv/snowball sampling and voluntarily chose to qualitatively answer a single open-ended question about their needs. All written responses were obtained *via* the internet; thus, responses could not be reliably tied to a specific healthcare visit or a type of indicator. These responses were coded reliably; however, it is not known the degree to which other patients in the sample also experienced IB or medical trauma but did not express this in their qualitative responses. In future research, each component of the BITTEN model needs to be assessed using standardized measures. Given that current participants voluntarily selected to participate, care should also be taken before generalizing these findings to the larger EDS patient population.

Conclusions and implications

Nonetheless, these findings suggest that HCPs may require additional training on how to care for and manage patients with complex and/or rare, hard to diagnose conditions. Results also highlight the need to attend to past negative healthcare-related experiences among vulnerable patient populations to break down patient-provider communication barriers and achieve higher quality healthcare. These experiences often include a history of institutional betrayal, medical or other traumas, and/or disrupted patient-provider trust. Each of these factors can directly influence patients' emotional health and their healthcare engagement behaviors,[16] as well as providers' perceptions of and approach to the patient. For example, a patient with an IB history might delay treatment seeking, withhold health-related information, not comply with treatment regimens, or behave in other ways providers may perceive as 'difficult.' BITTEN offers HCPs a framework for navigating these 'difficult' healthcare encounters in a trauma-informed, patient-centered manner. For example, by recognizing that an EDS patient is at increased risk for anger/irritability related to an undisclosed IB and/or trauma history, the provider may be more able to respond with empathy and mindfulness to this challenging patient presentation, subsequently helping to restore patient-provider trust.

These findings also point to a need for HCPs to use an interdisciplinary team to address the psychological aspects of care, as well as the structure of care, when treating patients with rare diseases. Furthermore, results suggest that many sufferers of EDS may already feel betrayed by their doctors, their healthcare system, and/or their own bodies, emotional responses, and support networks. They may feel lost in their diagnosis (or lack thereof), concerned about poor dissemination of EDS-specific information, and ultimately, distrustful of their HCPs and the healthcare institutions in which they practice. These concerns then map onto a series of negative expectations for future healthcare interactions as well as an increased likelihood of irritable and angry responses to current provider care. Instead of automatically considering these patients as 'difficult' and/or non-compliant,[17] current results suggest that providers should be moving from a frame of 'what is wrong with this patient' to 'what has happened to this patient?'.[18] Using this trauma-informed approach may also decrease provider burnout, while encouraging patient-centered encounters and greater quality of care. Furthermore, although thoroughly addressing patients' needs may seem difficult in the absence of time, resources, and available integrated care, many EDS patients may benefit from simple reassurance, attuned listening, and symptom validation. Efforts may also need to be directed toward EDS patients to facilitate their ability to have a stronger voice in their care processes and treatment options. Finally, system level change is required to increase referrals to evidence-based chronic pain management and support services, as well as to offer greater access to providers with specialized education about rare diseases.

Author Contributions

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Conflict of interest statement

The authors declare that there is no conflict of interest.

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Ethics statement

This study was approved by the Institutional Review Board at the University of South Alabama (approval number #1235568), informed consent was obtained electronically; participation was voluntary and anonymous.

Supplemental material

Supplemental material for this article is available online.

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