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Medical traumatic stress in cystic fibrosis: A qualitative analysis

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ABSTRACT

Background: People living with cystic fibrosis (PwCF) face a lifetime of potentially traumatic illness-related experiences that can lead to posttraumatic stress symptoms. Existing criteria for this type of post-traumatic stress, called medical traumatic stress (MTS), may not fully capture the CF experience. In this study we aimed to explore: 1) illness-related experiences perceived as traumatic in the setting of CF, 2) perceived MTS symptoms in PwCF, and 3) perceived health-related functional impairments from MTS.

Methods: Informed by our aims, we developed and piloted guides for semi-structured interviews and focus groups with PwCF, family members of PwCF, and CF medical providers. We then conducted a series of interviews and focus groups. The qualitative analytical process followed Deterding and Waters' three stages of flexible coding for in-depth interviews, generating key themes and sub-themes in each domain of study inquiry.

Results: We recruited 51 participants, including 24 PwCF, 7 family members of PwCF, and 20 CF care team members. Illness-related experiences perceived as traumatic were often characterized by themes of loss of agency, threats of bodily harm, and shifts in identity. Prominent MTS symptoms included shame, survivor guilt, burden guilt, germophobia, and symptom panic. Health-related themes of functional impairments perceived to result from MTS included poor adherence and strained relationships between providers and patients/families.

Conclusions: This is the first study to explore the specific experiences of MTS in PwCF. It highlights the need for screening that includes these specific exposure types and symptoms, which may be mitigatable with medical trauma-focused interventions.

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1. Introduction

People living with cystic fibrosis (PwCF) face a lifetime of illness-related experiences, including those within the medical system (e.g. medical visits, laboratory monitoring, invasive procedures, hospitalizations, and surgeries) [1] and beyond (e.g. lifestyle restrictions, emotional vulnerabilities, and illness-related uncertainties) [2].

Some illness-related experiences can be traumatic, leading to lasting psychological and physiologic symptoms of posttraumatic stress [3]. This type of posttraumatic stress, termed Medical Trau-

matic Stress (MTS), is defined by the National Child Traumatic Stress Network as "psychological and physiological responses of children and their families to pain, injury, serious illness, medical procedures, and invasive or frightening treatment experiences [4]," with similar constructs in adult populations [5]. While not a DSM diagnosis in itself, symptoms of MTS are thought to be primarily those of Posttraumatic Stress Disorder - intrusion, negative mood, hyperarousal, and avoidance [10,11]. MTS has been associated with decreased medication adherence [6] and reduced quality of life [7] in cancer and traumatic injury patients. In PwCF, the potential contribution of MTS to increased mental health [8] and medical burden is underexplored.

To elucidate the association of MTS with health outcomes in PwCF, we need patient-centered measurement tools for MTS in chronic illness patients - a population in which there is a lack of consensus surrounding MTS assessment [9]. We postulate that PwCF may have a distinct experience of MTS due to 1) repeated

Abbreviations: CF, Cystic Fibrosis; MTS, Medical Traumatic Stress; PwCF, People with Cystic Fibrosis.

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Table 1
Summary of interview guides.

	Primary Question	Suggested Areas for Additional Probing
People with Cystic Fibrosis and Family Members	Have (you/your child/spouse) had a medical experience that was traumatic for you/them?	What made the experience traumatic? What could have made it less traumatic? Personal surrounding factors (severity of illness, state of mind, previous medical experiences) Outside surrounding factors (family, friends, community, physical environment, medical system and personnel)
	How did this experience impact your/their life after the event itself was over?	Effect on patient's emotions (depression, anxiety, sleep, jumpiness/easy startle) Effect on patient's behavior (day-to-day life, relationships with others, participation in medical care)
Care Teams	What triggers medical trauma in patients with cystic fibrosis?	Consider: events they undergo that might be scary or make them feel their life is threatened, things done to their bodies, interactions with medical providers. Consider important moments in the illness journey for the patient: diagnosis, sharing diagnosis with others, daily experience of the illness.
	What symptoms do patients have when they are experiencing trauma from a medical event?	How may the trauma impact: emotions, behavior, physical wellbeing?
	What is the impact of those symptoms?	Effect on patient's behavior (day-to-day life, relationships with others, participation in medical care, ability to achieve goals)

potentially scary and life-threatening medical events 2) lifelong, incurable illness with inherent life-threat and 3) stressful daily health management burdens.

In this qualitative study, we engaged PwCF, family members of PwCF, and CF care team members in interviews and focus groups to explore: 1) illness-related experiences perceived as traumatic in PwCF, 2) MTS symptoms prominent in PwCF, and 3) perceived health-related functional impairments from MTS. This multi-stakeholder approach centers the experience of PwCF while leveraging the external perspective of family members and CF care teams to capture a full view of the experience of MTS in PwCF, and to facilitate exploration of parallelism and divergence between stakeholder groups.

2. Methods

2.1. Setting and participants

We recruited a convenience sample of participants nationally, January - June 2021, through CF listservs (CF Foundation's Mental Health Listserv, Registered Dietician Listserv, Social Work Listserv), social media (Facebook, Twitter), targeted provider outreach, and the CF Foundation's Community Voices Program. Eligible participants included: 1) English or Spanish-speaking PwCF and their family members ≥ 10 years and 2) CF providers with ≥ 1 year of direct CF patient care experience. Planned enrollment was for at least 20 interviews and 2-4 focus groups, with sample extension if needed to achieve thematic saturation and to capture diverse perspectives [12]. We used iterative sampling, whereby thematic analysis occurred in tandem with data collection to determine the saturation point and inform the final sample size. The minimal racial and ethnic diversity of our initial sample prompted purposive sampling to increase enrollment of diverse participants.

This study was approved by the institutional review board at the University of California, San Francisco 20-31965. Prior to participation, participants were provided with a summary of our study objectives, and participants ≥ 18 years old provided written informed consent. For those ≤ 18 , we obtained parental consent and participant assent. Our research team included the authors and a steering committee (supplemental table 1).

Study interactions occurred virtually over a HIPPA-secure teleconferencing platform (Zoom) and were recorded. Recordings were transcribed (transcriptionwing.com). Dedoose qualitative software was used for coding and analysis.

PwCF and family members participated in semi-structured interviews due to the sensitive nature of the topics and the format's

allowance for participant expression with guidance [13]. Care team members participated in focus groups to enhance the validity of interview findings and facilitate interactional discussion to extrapolate complex details [14]. No repeat interviews were conducted, and study participants were not re-contacted to review transcripts or provide feedback on findings.

2.2. Data collection instruments

Guided by the domains of inquiry, we developed interview guides covering: 1) perceived traumatic illness-related experiences 2) MTS symptoms, and 3) functional impairments. Each interview guide was tailored to the pertinent stakeholder group. We developed suggested areas for additional probing informed by the biopsychosocial model, criteria for traumatic stress disorders [10], and medical traumatic stress frameworks [4,11] (Table 1). We piloted the interview guide in three PwCF prior to use for this study. Demographics were self-reported.

2.3. Analysis

The content analysis followed Deterding and Waters' three stages of flexible coding for in-depth interviews. This top-down approach entails broad categorization data at the onset of analysis using "index codes," followed by a constructive, iterative process of theory development based on cross-interview analysis within each index code [15]. In stage one, we indexed data from each interview under three index codes: "Traumatic Illness Related Experiences," "Medical Traumatic Stress Symptoms," and "Functional Impairments." During this, we generated memos and began to develop analytic codes. In stage 2, we engaged in cross-interview comparisons within index codes, iteratively refining the analytic codes into central themes and subthemes. In stage 3 we re-evaluated the themes and subthemes with input from the steering committee, verifying each and considering alternatives.

3. Results

We conducted 31 interviews and three focus groups, for a total of 51 participants (Table 2). One provider signed a consent but was unable to attend the focus group. No other participants dropped out. Interviews averaged 79 minutes (range 39-148 minutes). The three focus groups included 8, 10, and 3 participants, averaging 83 minutes (range 61 to 96 minutes).

Table 2

Participant demographics (n=51).

	PwCF/Family Members (n=31)	CF Care Team Members (n=20)
Age in years (median, IQR)	31 (27-43)	39 (33-42)
Female gender	21 (67%)	19 (95%)
Stakeholder Type		
Person with CF	24 (77%)	-
Adult Person with CF	21 (68%)	-
Child/Adolescent Person with CF	3 (10%)	-
^a Parent or Spouse of a Person with CF	7 (22%)	-
Care Team Member		
Medical doctor	-	7 (35%)
Social worker/Psychologist	1 (3%)	7 (35%)
Dietician	-	4 (20%)
^b Other provider	-	4 (20%)
^c Years endured with cystic fibrosis diagnosis (median, IQR)	26 (15-36)	-
Years of caring for cystic fibrosis patients (median, IQR)	-	6 (4-13)
Region of the United States		
Northeast	7 (23%)	3 (15%)
Midwest	4 (13%)	6 (30%)
South	13 (41%)	3 (15%)
West	7 (23%)	8 (40%)
Race		
White	29 (94%)	12 (60%)
Asian	-	8 (40%)
Prefer not to respond	2 (6%)	-
Ethnicity		
Hispanic or Latino	4 (13%)	1 (5%)
Insurance Type		
Private	16 (52%)	-
Government sponsored or none	15 (48%)	-
Highest level of education		
Grade 12 or below	6 (19%)	-
College with or without advanced degree	25 (81%)	-

^a Family members included two parents of PwCF under 5 years, three parents of teenage PwCF, and one spouse of a PwCF. One parent of a PwCF was also a member of a CF care team, and they were interviewed individually given this unique positionality.

^b Other provider includes: respiratory therapist (n=2), registered nurse (n=1), pharmacist (n=1).

^c Years since diagnosis of individual with CF or parent/spouse's loved one with CF.

PwCF's ages ranged from 11 to 61 years (median 34). Family members' ages ranged 25 to 55 years (median 38). Two parent/child dyads were interviewed. PwCF and family members had extensive lived experience, enduring a median of 26 years with a personal or family member's CF diagnosis (IQR 15-36 years). CF care team members held a wide range of professional roles, represented all U.S. regions, and had a median of 6 years caring for PwCF (IQR 4-13 years) (Table 2).

Thematic analysis is outlined in Fig. 1. Table 3 shows representative quotes of key subthemes. In the text below, patient and family member quotes are prefixed with "PF," and care team member quotes are prefixed with "CT."

3.1. Perceived traumatic illness-related experiences

Participants reported a breadth of potentially traumatic illness-related experiences, including daily stressors, traumatic life events, and chronic stresses. These experiences spanned all stages of development, from early childhood illness experiences to adult CF diagnoses. Adolescence and young adulthood were commonly identified as times of worsening physical illness and an increase in traumatic illness-related experiences.

Across stakeholder groups, illness-related experiences perceived as traumatic shared common characteristics captured in three themes: 1) threat of bodily harm, 2) loss of agency, and 3) triggered shifts in identity.

3.1.1. Bodily harm

Perceived threat of bodily harm was noted in the majority of interviews with PwCF and family members. Two key subthemes emerged. *Perceived life threat* was reported both in the medical setting, such as with acute clinical decompensation (CT1), and in non-medical settings, such as learning of the life-limiting nature of CF while doing independent research at home or in a library (PF1). *Painful medical experiences* captured a wide spectrum of experiences within the medical system, from blood draws and sputum cultures to intensive care admissions and major surgeries (PF2; CT2).

3.1.2. Loss of agency

Among PwCF and family members, nearly all identified a traumatic illness-related experience that included loss of agency, characterized by disempowerment or lack of control. One major subtheme, *perceived lack of control during medical encounters* (PF3, CT3), was seen in the context of hospitalizations, medical procedures, and provider interactions. A second was *lack of control over their health and illness trajectory*, wherein PwCF and their family members commonly noted the looming threat of sudden, unpredictable, and uncontrollable illness progression (PF 4, CT4). A third subtheme, *lack of control over systems* (PF5, CT5), was particularly emphasized by care team members who highlighted the role of healthcare systems that limited patient access to some types of care. Examples included insurance denials for potentially life-saving medications and difficult to navigate communication systems in doctor's offices that made it challenging to reach a provider for advice.

3.1.3. Triggered shifts in identity

Half of PwCF and family members identified traumatic illness-related experiences that triggered shifts in their identities – either from that of a "well" to a "sick" person (PF6, CT6) or from a "sick" to a "well" person (PF7, CT7). Circumstances leading to identity shifts were heterogeneous. Examples include the death of a peer leading to feelings of increased personal vulnerability; a clinical deterioration; or, conversely, access to potentially lifesaving treatments like lung transplant or CF transmembrane conductance regulators. In the latter cases, examples of feeling surrounding the shift from a "sick" to "well" identity included: overwhelm from the sudden need to financially plan for a longer lifespan, a lost sense of purpose, and uncertainty about how to interact with others who had not benefitted similarly.

3.2. Medical traumatic stress symptoms

Participants identified MTS symptoms within the DSM-5 domains of intrusion, avoidance, negative mood, and reactivity. Key subthemes highlighted prominent manifestations of these symptom domains within the context of CF (Table 3). PwCF and their family members noted symptoms lasting from months to years after the traumatic illness-related experience.

3.2.1. Intrusion

When describing intrusion symptoms, *flashbacks* (PF8) and *scary dreams* (PF9) were emphasized by PwCF and family members, especially after stays in intensive care.

3.2.2. Avoidance

Symptoms pertained to avoidance of medical care, such as *dreading* (PF10, CT10) and *avoiding* interactions with the healthcare system (PF11, CT11), as well as *avoidance of social interactions* (PF12, CT12). Care team members emphasized avoidance symptoms, reporting both less frequent visits and lack of engagement from patients with traumatic illness-related experiences.

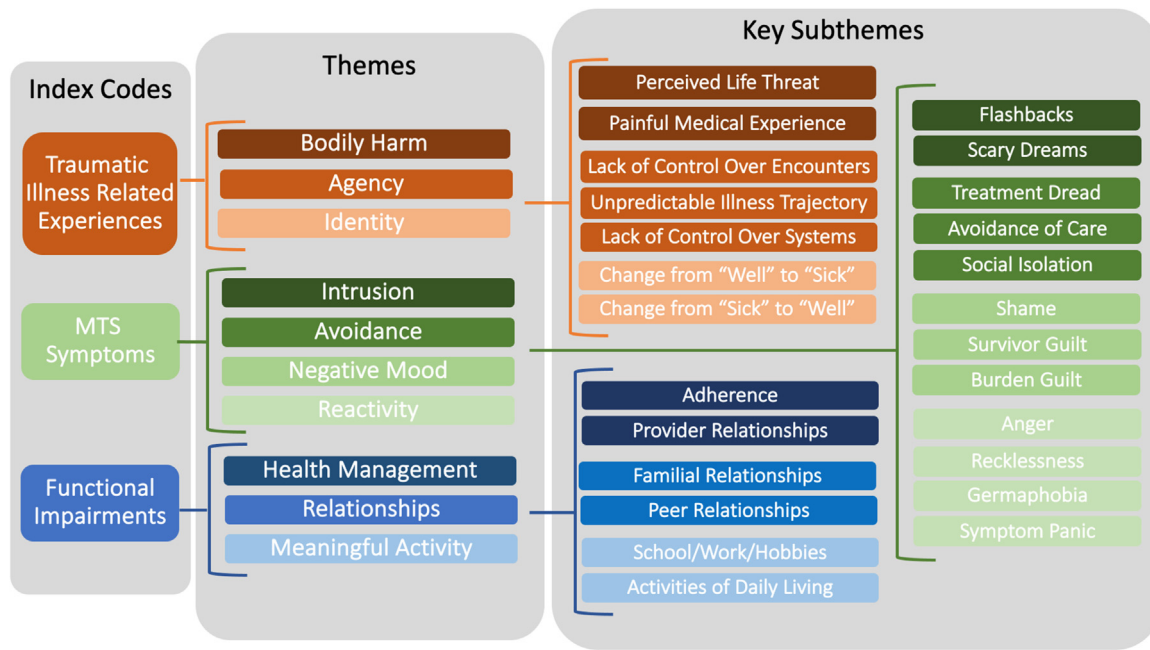


Fig. 1. Overview of qualitative analysis.

3.2.3. Negative mood

Within this domain, symptoms reported included depression, persistent and exaggerated negative beliefs, and diminished interest in activities. Particularly prominent were sub-themes of *shame* (PF13, CT13), *survivor guilt* (PF14), and *burden guilt* (PF15). Included with *shame* was a sense of embarrassment due to the illness's effect on PwCF's bodies, reported in different contexts across the lifetime including school, dating, parenting, work, and in the medical setting.

Survivor guilt, a term commonly named by PwCF, referred to feelings of guilt that occurred when benefitting from a treatment that others did not have the opportunity to enjoy. Examples included: guilt that they survived when a lung donor did not; guilt about benefitting from breakthrough CF treatments when others are ineligible; and guilt that they continued to live when other PwCF died.

Burden guilt captured guilt associated with the impact that a PwCF's illness had on those around them, including parents, spouses, children, friends, and society.

3.2.4. Physiologic arousal

PwCF and their family members reported experiencing *anger* (PF16, CT16), *recklessness* (PF17), *germaphobia* (PF18, CT18), and *symptom panic* (PF19, CT19). *Anger*, often that felt uncontrollable, could be directed at oneself, family members, friends, healthcare providers, and healthcare systems. Care team members noted that anger was at times associated in the clinical setting with tension between patients, families, and care team members.

Recklessness, discussed by PwCF but not care team members, was particularly noted as a symptom in adolescence and young adulthood; it included risky behaviors both related to and beyond the scope of medical care, such as drug use. *Germaphobia* was noted by many participants and exacerbated by the COVID-19 pandemic.

Symptom panic occurred when small or slight symptoms drew patient fixation and caused physiologic responses including sweating, fast heart rate, and shortness of breath. This was particularly noted when the perceived traumatic illness-related experience in-

involved a threat of bodily harm, like a severe pulmonary hemorrhage.

3.3. Functional impairments

3.3.1. Impaired health management

The impact of MTS symptoms on health management was captured by two subthemes, 1) *impaired adherence to treatment regimens* (PF20, CT20) and 2) *impaired relationships with medical providers* (PF21, CT21). Reported impact on adherence included decreased adherence to daily medication and respiratory therapy regimens, as well as impaired participation in routine and urgent medical visits. The impairment of relationships with medical providers was endorsed by all stakeholder groups. Participants linked MTS symptoms to decreased quality of therapeutic relationships due to communication challenges and erosion of trust.

3.3.2. Impaired relationships

The impact of MTS symptoms on key *familial* (PF22, CT22) and *peer* (PF23, CT23) relationships were reported by PwCF and their family members. Manifestations of these impairments varied based on social situation, developmental stage, and health context. Examples included distanced relationships with parents due to fear of abandonment; unwillingness to engage in peer relationships due to shame and a desire to hide diagnosis; and estrangement from siblings due to survivor and burden guilt.

3.3.3. Impaired involvement in meaningful activities

Impaired involvement in meaningful activities was also present across developmental stages. It was highlighted as *impaired academic performance* (PF24, CT24) in children and adolescents, and *impaired ability to perform work activities* in adults due to episodes of symptom panic or flashbacks (PF25).

3.4. Convergence/divergence and special populations

Overall, there was close convergence on the themes and sub-themes between PwCF, family members, and care teams. Notable

Table 3
Example quotes by theme and subtheme.

Index Code	Theme	Key Subtheme	Example quote from a person with CF or family member	Example quote from CF Care team members	
Traumatic Illness Event	Bodily Harm	Perceived Life Threat	PF1: "It was very disheartening to read my life expectancy was 19 years old, and I was 19 at the time. That was traumatic. Without the internet your whole life, that was the first time. My parents never told me that, that I'd have a shortened lifespan."	CT1: "Coughing up blood can be really scary...I had a patient who was hiking once...it was a hot day, and he didn't have enough salt or he was imbalanced, and he ended up passing out and had to be like med-evacuated off a hiking trail like in front of his friend, and that was really traumatic."	
		Painful Medical Experience	PF2: "Then I went to the ER. I've been to the ER quite a few times before that too. So, that wasn't that traumatic in and of itself, but then just laying on the bed...the physician, and then he had like three...students that were watching the procedure. I remember I was obviously awake. I was just turning my head to the other side, just trying to take deep breaths, breathing through it. It was pretty painful. Then I remember kind of like wincing, like making a moaning sound, ... and one of the students I remember asked [the doctor]. He was like, 'It looks like it's hurting her. Does it need more lidocaine or some sort of like the numbing stuff?' He [the doctore] is like, 'No...only when they like scream and their eyes pop open, that's when you know it's like a level of like really, really painful and she's not doing that. So, it's fine.'" PF3: "I never felt like I had a choice to get up and leave or to say, 'Hey, we've got to stop' or anything like that. I never felt that choice."	CT2: "NG tubes is a huge one [traumatic experience]... Enemas, IV starts, accessing a port is one...getting a PICC line could be very stressful...."	
		Agency	Lack of Control Over Encounters	PF4: "When the bacteria were inside my lungs, I wasn't feeling any different. I felt completely fine. But then when I went to go do a PFT they're like, 'Oh, my god. Your lungs are really bad right now.' I was like, 'Oh, I didn't know that.' Then further [on] they're like, 'You have really bad bacteria.' I'm constantly worried about [the bacteria] and, currently, I feel fine but I could be dying on the inside." PF5: "I just felt so abandoned at this new center. I just was disgusted because I had left such good care and I got treated this way and it was traumatizing...It's not like just one clinic has all these issues, it's many places and patients suffer and I know I'm not the only one too. I hear it in my group all the time of other patients who have horrible experiences or are forgotten or have to make each repeated call. So, it's just exhausting, depressing; it's heartbreaking."	CT3: "I think outpatient and inpatient, any time that there needs to be a poke...they will take a bunch of tries, and then...they hold the child down or get the parent involved to hold the child down, and then it becomes really traumatic for them." CT4: "You know, over time, it's going to progress...with some admissions, people may wonder, 'Is this the worst thing on my disease? Is this going to be a new baseline? What does this mean for me, like the long-term impact?' I think that that ... itself may be...triggering."
	Identity	Unpredictable Illness Trajectory	Lack of Control Over Systems	PF6: "I'd been coughing out mucus all the time. I'd been having digestive issues, but it wasn't until there was a name for it, and I realized that I was labeled. In that moment, my opinion of myself changed."	CT5: "...Mixed messages, not being able to get their treatments from multiple pharmacies because of insurance issues...not having access or not being able to afford [the treatment] can be traumatic."
			Change from "Well" to "Sick"	PF7: "...So it just feels like I've been trying to survive my whole life. Now, I'm on Trikafta and I'm recognizing like, 'Wow, I no longer have to define myself as a sick person.' It's been challenging because I don't know who I am outside of that."	CT6: "Lot of adults tell me that it still replays in their head the first time they ever learned about their diagnosis or learned about their limited life expectancy, and that continues to be a pattern in their head throughout their adult care as well." CT7: "We've also seen a lot of opposite from the modulator trauma where you would think that they're excited about this great improvement but they now actually have to think about their future and what they're going to do to actually take care of themselves when their parents are gone; things that they've just never thought about."
		Agency	Change from "Sick" to "Well"		

(continued on next page)

Table 3 (continued)

Index Code	Theme	Key Subtheme	Example quote from a person with CF or family member	Example quote from CF Care team members
MTS Symptoms	Intrusion	Flashbacks	PF8: "It has led to moments of having actual flashbacks that were triggered by someone just putting a hand on my shoulder and me remembering being pushed into my [hospital] bed."	
		Scary Dreams	PF9: "After the transplant, anytime I closed my eyes, I had real bad nightmares, just vivid, visceral, violent things."	
	Avoidance	Treatment Dread	PF10: "Currently, with a PFT, it does start days before. I just get a little anxious, a little bit on edge for maybe three or four days before."	CT10: "I've had a patient who literally would shake talking about her going for lab work, just talking about it, so she'd have a physical response to the stress of the idea."
		Avoidance of Care	PF11: "I would physically try to escape...one time I got into the elevator before they could catch me when they were trying to get me [to do] bloodwork and I... just tried to run away."	CT11: "...This kind of fear of going to the hospital after having a significant near-death experience. Like if they have a pneumothorax...they don't want to come near us anymore basically."
		Social Isolation	PF12: "I think it's made me feel like I need to just do these things alone. Maybe I push people away when I'm hurting or I'm not well, maybe out of the fear that they were going to make it worse."	CT12: "[Patients] just getting into a dark pit type of thinking, which then we see kids don't want to move out, they don't want to go to school, they don't want to be in relationships."
	Negative Mood	Shame	PF13: "I didn't want anyone to know. I was embarrassed, because it was so disgusting...shame was the main feeling, the main emotion that I had at that point."	CT13: "[I] had a couple of my patients say that they feel bad for having to be on Medicaid even though they're more than able to work."
		Survivor Guilt	PF14: "[I] was just thinking like, 'How could I dare to be suicidal when this person gave their life?' and then that made me more depressed and more suicidal."	
		Burden Guilt	PF15: "I was really worried about how other people were feeling and how they were perceiving my health issues and what they thought was going to happen. I didn't want anything to happen to me because I didn't want anything to happen to them, and for them to go through something they don't deserve. If I were to die, I wouldn't want all these people to be crushed and hurt. That's one of my biggest fears."	
	Reactivity	Anger	PF16: " [I] wake up just like I'm furious for no reason, right, or with good reason, but it wouldn't take any situation. Just opening my eyes in the morning, and already, I could just like feel it coursing in my veins. I hated everything. I hated everyone. Everything set me off. That's all I felt, was just anger every single day."	CT16: "Anger is really common, which then really interferes with your ability to partner around care."
		Recklessness	PF17: "I was drinking alcohol, smoking cigarettes, smoking pot, having sex, getting in fights, talking back to teachers, getting in trouble, all that stuff because I realized, if I'm going to die, all the things you're supposed to do and most people do in college and this and that, I was like I'm not going to have that opportunity then. So, let's get this out of the way. If you're going out, go out in a blaze of glory."	
		Germaphobia	PF18: "My house is very clean. [My daughter] was little when I found out she had CF, I cut bath toys in halves because I wanted to see which ones held the moisture well and which ones didn't."	CT18: "We have a patient [whose]... germaphobia has gone through the roof over the past few years and especially with COVID, and she went from being in school to 100% homeschooled, to needing her brother to be homeschooled, to now having to go into an inpatient OCD program. I mean, I think it definitely obviously impacted her ability to function"
		Symptom Panic	PF19: "If [my lung] is collapsed, am I going to be somewhere where I'm going to safe? Am I going to be able to get care that I need? It's going to happen really suddenly this time. Is it going to happen in a matter of minutes? Then like, who's going to help me get to the hospital? It was like all these what-if scenarios and like playing those out and like how I would react to what I would do."	CT19: "I have a few kids who have said that when they get a coughing fit, whether it's productive or not, sometimes they feel like they can't catch their breath, and the more they cough, the more that they need to cough, and they're worried that someday they just won't be able to breath anymore and will die."

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Table 3 (continued)

Index Code	Theme	Key Subtheme	Example quote from a person with CF or family member	Example quote from CF Care team members
Functional Impairments	Health Management	Adherence	PF20: "I would just think, '...If I don't take that medication, I can actually go out and have some fun. I can actually enjoy my life.' So, for me, I was thinking, 'I'm trying to enjoy my life and, yes, it led me into this cycle of no enjoyment and the cycle of hating myself for not taking care of myself.' I had a lot of self-loathing, a lot of guilt, and yet, once I get out of the hospital, I just do it all over again."	CT20: "I think they [patients] get angry, and so they just stop doing their care. They are frustrated by us and the way to get back is, 'I'm not going to do my vest. I'm not going to take my pills.'"
		Provider Relationships	PF21: "I literally wanted to grab her by her throat and scream at her and tell her, 'If you tell me to give up one more time, this is not going to end well,' because...the fear and the anxiety turned into anger really quickly." PF22: "I just screamed at them [parents] and say, 'It's your fault that I even have this disease. It's genetics,' just these terrible things. So, that created this huge rift in our relationship."	CT21: "We do talk about options and then some parents express how they don't want to hear it again, [they say] like 'I don't want to see the person who brought it up again' and it starts to get emotional." CT22: "[Patients] definitely distance themselves from their parents, whether it's physical or psychological...if you have a parent who's not responsive to you in a time of need like medical trauma, it can build a big resentment around your disease also like, 'Well, if I didn't have my CF, then my relationship with my parent might be different.'"
	Relationships	Familial Relationships	PF23: "I couldn't open up to people. I was hiding. I was ashamed...I was too embarrassed to take all these pills in front of people, because I didn't want them to ask what's going on. I didn't want to have to explain my illness."	CT23: "Going through the scary traumatic experience or life threatening condition maybe feels isolating. They cannot relate to their friends who don't have that kind of experience. That could worsen the relationships...I can think of several patients who sort of just hole up in their rooms."
	Meaningful Activity	School, Work, Hobbies	PF24: "I remember like I would sit in my classrooms, and like we were taking a test, and I would have so much anxiety that something terrible was going to happen to me. At that time, I hadn't had any lung collapses yet, but I was just terrified like there's something, like I was going to stop breathing, that something bad was going to happen to me while I was like taking a test, and I wouldn't be able to like leave the room and I would like have all this attention called to me." PF25: "If I heard somebody sneeze or cough in the store, no matter what I'm supposed to be getting or buying, I had to leave the store."	CT24: " I had one patient who developed kind of almost germaphobia going to school, and had school avoidance, because she was so worried about getting sick from the playground. "
		Activities of Daily Living		

areas of divergence included lack of care team comment on: intrusion (including flashbacks and scary dreams), survivor and burden guilt, recklessness, and impaired activities of daily living.

Subgroup analysis of the four Latino(a) participants revealed that late diagnoses and misdiagnoses contributed to MTS in these patients. Racial biases were perceived to impact care in these participants and contributed to perceived threat of bodily harm or loss of control. There was downstream impact of these events on communication with and trust in healthcare teams.

4. Discussion

This study is the first to incorporate stakeholder perspectives on the CF experience of medical trauma. Illness-related experiences perceived as traumatic varied from discrete medical events to chronic and daily stressors. They occurred within and outside of the medical setting. Traumatic illness-related experiences often involved loss of agency, the threat of bodily harm, and shifts in identity.

PwCF reported symptoms classically described within the DSM-V posttraumatic stress symptom domains, but they also reported symptoms more particular to chronic illness: shame, survivor guilt, burden guilt, germaphobia, and symptom panic. Participants iden-

tified medical functional impairments from these symptoms, including alterations in health behaviors and patient/provider relationships. These findings emphasize that optimization of health and wellness for PwCF should include MTS screening, mitigation, and treatment [16].

The illness-related experiences in the medical setting that our participants identified – including surgeries, laboratory draws, and shots – have long been recognized as sources of anxiety and trauma for children [11], and recently recognized amongst PwCF [17]. Our study highlights that MTS can also be triggered by illness-related experiences occurring outside of the medical setting. In both settings, perceived traumatic illness-related experiences were united not by type of event, but rather the individual's appraisal of those experiences as entailing threats of bodily harm, loss of agency, and/or triggering identity shifts. These characteristics closely mirror features of other traumatic stressors [10,18].

We identified symptoms of MTS that are minimally discussed in previous MTS and CF literature and warrant further investigation, including survivor and burden guilt, anger, germaphobia, and symptom panic. Previous discussion of survival guilt has largely focused on parents of PwCF and siblings [19]; however, in the current era of highly effective modulator therapies, it appears from PwCF as well [20].

The themes of survivor guilt, flashbacks, scary dreams, recklessness, and burden guilt were prominent in interviews with PwCF, but not mentioned by care team members. The lack of provider attention to these symptoms raises concern that care teams may not be aware of or screening for them. Fear of judgment could potentially decrease PwCF's willingness to share these symptoms [21].

Stakeholders identified health-related functional impairments associated with MTS symptoms. Difficulty adhering to treatment regimens and strained relationships with providers are known challenges for PwCF and their care teams [22]. Investigation is needed into the contribution of MTS to these challenges. The potential of relationship-centered communication trainings to mitigate MTS should also be explored [23].

Our sample size was insufficient to stratify themes by demographics. While our recruitment methods included explicit strategies for recruiting Black, Indigenous, and People of Color, our final sample had limited racial and lingual diversity. Despite advances in CF care and treatment, minorities continue to suffer the greatest burden of CF disease, including modulator ineligibility [24], which was a potentially traumatic illness-related experience for our participants and can have profound psychosocial impacts [25]. Additionally, these patients may be at risk for concomitant MTS and race-based traumatic stress, given the pervasive individual, institutional and structural racism within the United States healthcare system. Future work exploring the intersection of MTS and race-based traumatic stress is imperative.

The qualitative methodologies we employed are subject to recall bias and influence from the positionalities of the research team [26], (supplemental table 1). Focus group data can also be skewed by dominant voices and group think [27]. As generalizability from qualitative studies is limited, any future screening measures based on our findings should be tested prospectively and validated in a larger, more diverse population.

Our study has important implications for the field of MTS and the care of patients with pediatric-onset chronic illnesses. The MTS symptom themes described largely fall under the DSM-V criteria for posttraumatic stress disorder, [10] and therefore are identifiable with existing questionnaires. However, current screeners lack specificity for the chronic illness context that our sub-themes capture. We recommend that future screening for MTS in PwCF should: 1) identify experiences within and beyond the medical system, 2) evaluate for medical experiences appraised to represent threats of bodily harm, loss of agency, and triggered shifts in identity, instead of specific procedure types, and 3) assess illness-specific MTS symptoms. Next steps in research include: 1) development of a theoretical framework capturing MTS in chronic illness, 2) creation and validation of a MTS of chronic illness screening measure, and 3) rigorous evaluation MTS interventions in chronic illness populations, including the impact of expanded mental health access and integration of mental health experts into clinical care teams.

Conclusion

This is the first study to explore the experience of MTS in PwCF. Our findings highlight the need for future research into targeted approaches to MTS for PwCF, including the development of screeners that capture the characteristics of illness-related experiences and MTS symptoms identified here.

CRedit authorship contribution statement

Addison A. Cuneo, MD: Conceptualization, Methodology, Project administration, Formal analysis, Investigation, Data curation, Writing – original draft, review, and editing, Visualization, Funding acquisition. **Simon Outram, PhD:** Methodology, Formal

analysis, Investigation, Resources, Data curation, Writing – review and editing. **Meghan L. Marsac, PhD:** Methodology, Writing – review and editing, Supervision. **Siena Vendlinksj:** Resources, Writing – review and editing, Visualization, and Project administration. **Melvin B. Heyman, MD:** Methodology, Writing – review and editing, Supervision. **Ngoc Ly, MD, MPH:** Methodology, Writing – review and editing, Supervision. **Emily von Scheven, MD, MAS:** Methodology, Writing – review and editing, Supervision. **Emily R. Perito, MD:** Conceptualization, Methodology, Writing – review and editing, Supervision.

Declaration of Competing Interest

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Supplementary materials

Supplementary material associated with this article can be found, in the online version, at doi:10.1016/j.jcf.2022.12.008.

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