Abstract

Background. Collectively, chronic inflammatory diseases take a great toll on individuals and society in terms of participation restrictions, quality of life, and economic costs. Although many qualitative studies have reported patients' experiences and challenges living with inflammatory diseases, they are sometimes criticized as non-transferable due to small and highly specific samples. Larger-scale studies could verify patient perspectives across a broader sample and examine similarities in the patient experience across different types of inflammatory diseases.

Objective. To identify the significant consequences of inflammatory arthritis, psoriasis, and inflammatory bowel diseases on daily life and explore commonalities across diseases.

Method. A cross-sectional online survey was designed by patient research partners and distributed by representatives of patient organizations via their social media channels and a newspaper story. One open-ended item asked about burdens and responsibilities experienced in daily life. Informed by narrative traditions in qualitative health research, we applied an iterative content analysis to participants' written accounts in response to this item. This approach is an example of a study conceived, conducted, and interpreted with patients as research partners.

Results. 636 Canadians submitted surveys, median age band 55-64 years, 80% women. 540 wrote substantive responses to the open-ended item. Four main narratives were identified: (1) daily life disrupted; (2) socio-economic vulnerabilities; (3) stresses around visible, invisible, and hiding disabilities; and (4) actions aimed at staying positive. Ways in which participants experienced social stigma, pain and fatigue, balancing responsibilities, and worries about the future appeared throughout all four narratives.

Conclusion. People living with chronic inflammatory diseases affecting joints, skin, and digestive tract report important gaps between health, social, and economic support systems that create barriers to finding the services they need to sustain their health. Regardless of diagnosis, they report similar experiences navigating the consequences of lifelong conditions which has implications for policy makers. There is a need for outcomes measures in research and service delivery to address patient priorities, and for programs to fill gaps created by the artificial administrative separation of health services, social services, and income assistance. [335 words]

Key Words
Patient Partnership; chronic disease; inflammatory disease; patient experience; public engagement

Introduction

Patient engagement in health research has been building over the past two decades, with examples of effective collaborations between patients and researchers being reported with increasing frequency. The benefits of patient engagement across the research process include identifying research questions of greater relevance to patients' concerns, improved participant enrolment and retention rates, and knowledge translation strategies that are more readily understood or adopted by community members. [1] As part of multi-project research grant application, our research partners surveyed members of the public and patient groups to help justify research priorities. The purpose of this paper is to share the analysis of an opened-ended survey item probing the key issues faced by people living with chronic inflammatory diseases.

This paper focuses primarily on inflammatory arthritis, psoriasis, Crohn's disease, and ulcerative colitis. The patient partners (CK, AS, MA, GA) live with one or more of these diseases. All of these conditions are systemic in nature, range from mild to severe, and are characterized as episodic, meaning people live with the uncertainty of exacerbations and remissions either from the natural course of the disease or its medical management. Studies on the impact of living with these inflammatory conditions show activity disruption, [2 - 6] reduced productivity, [5 - 7] and high personal costs, threatening financial security. [2, 7] Among women with early rheumatoid arthritis, McDonald et al. found that uncertainty was particularly problematic, as women experienced good days (able to engage in typical routines and daily activities), bad days (experiencing limitations in typical routines and daily activities), and worse days (often halting usual activities due to pain, fatigue, or recovering from symptom flares). [8] Adapting to activity disruption threatened self-identity and sense of self. [8] Similar experiences have been reported by adults living with established

inflammatory bowel disease [9, 10] and psoriasis. [11 -13] Potentially counterbalancing these experiences is the reported sense of empowerment, confidence, and contribution to the greater good that arises from patients meaningfully engaging in the research process from inception to dissemination of findings. [14] In-depth descriptions of activity disruption are commonly reported in qualitative research, which by nature focuses on relatively small numbers of participants. These kinds of studies are valuable to patients because they corroborate their experiences, show they are not alone, and provide strategies for living well, interacting with health professionals, and advocating for resources. They are valuable to professionals for enhanced understanding of the impact of living with different diseases, placing patient experiences in context, and ultimately help improve patient-provider communication and more compassionate care. [15] Inviting a large number of people to respond to an open-ended question typical of qualitative studies may support the transferability of findings from small studies, and that opportunity arose in the present partnership. Box 1 describes the research context in which the present paper was situated.

Box 1. Research Setting

PRECISION is a pan-Canadian team of over 30 members, including patients and health professionals, who developed a successful funding application for a collection of inter-related research projects aimed at reducing complications from inflammatory diseases. [16] PRECISION is an acronym for the project title, PREventing Complications from Inflammatory Skin, joint, and bowel conditIONs, and the diseases under study include psoriasis, rheumatoid arthritis, lupus, ankylosing spondylitis, gout, Crohn's disease, and ulcerative colitis. The team came together in response to a national research initiative on chronic inflammation to foster collaboration across body systems or disease entities. PRECISION objectives include assessment of the risk and burden of complications and consequences of these diseases and the testing of novel health services aimed at preventing or mediating those complications. The patient partners in PRECISION are representatives of four consumer-oriented organizations specific to diseases affecting skin, joints, and bowel. PRECISION aims to produce research that addresses issues common across different chronic inflammatory disease groups, and it is from this mandate that this paper proceeds.

From the outset, patient partners shaped the research plan in collaboration with interdisciplinary researchers. They drew upon the experiences of their considerable constituencies by conducting an online survey to help justify study objectives important from the patient perspective. This survey was carried out by patients and researchers, for patients. The insight gained from the survey helped the PRECISION team to plan studies to better understand both the medical complications of inflammatory diseases, and the social and emotional consequences that are integrally tied to the provision of health care services and the patient-provider relationship. That the team was funded, with peer review commending the centrality of the patient perspective, is one indicator of the success of patient engagement. It is one portion of the patient survey that is described in the present paper.

Methods

A cross-sectional design was used and consent was implied by submitting a completed survey. Ethical approval was obtained from the Behavioural Ethics Review Board of the researchers' university.

Recruitment. Each of the patient partners is affiliated with a patient organization; these four organizations distributed the survey link to their members nationwide and connected with a newspaper reporter who wrote a brief story that included the survey link in the print version of a metropolitan daily newspaper and the reporter's blog. There were no explicit inclusion criteria other than the survey notice specifically invited people with inflammatory joint, skin, or bowel diseases to have a say in research and complete the survey anonymously. The survey was open for three weeks.

Survey Content. Patient partners co-designed an online survey with researchers on the PRECISION team. The partners vetted a large number of potential items to reduce the total number and ensure clarity of the retained items. In addition to basic demographic information (e.g., diagnosis, sex, age group in 10-year age bands, urban vs rural place of residence), the survey contained closed response and open-ended items to gather patient perspectives on medication use, knowledge about potential disease complications and

treatments/interventions, lifestyle habits (e.g., physical activity), and experiences living with inflammatory diseases. The responses to closed response items helped justify the grant application with respect to needs around specific diseases, complications, medications, and physical activity. [17] In the present paper, we focus on text responses to the open-ended question: *what are some of the burdens and responsibilities you face in managing or living well with your illness?* There was no word limit imposed on stories written in response to this item, nor was it required that participants enter any text.

Data Analysis. Responses were downloaded verbatim into an Excel file for tabulation (keeping text responses linked to demographic descriptors such as age and diagnosis) and analysis. The burdens and responsibility question generated numerous stories and commentary. Tallying was avoided because the spontaneous responses to the open-ended question meant that some respondents introduced new topics that, if tallied, would not represent the proportion of respondents who shared that view; counting was not found to yield specific or meaningful data. [18] Accordingly, we drew upon narrative traditions that privilege personal accounts and experiences to conduct a thematic content analysis of these text responses. [19] We sought to understand *what* people experienced rather than *how* they described it, making thematic content analysis more appropriate than other forms of narrative models for this dataset. [19] Thematic content analysis is suitable to participatory types of research because it is generally understandable by all audiences, highlights similarities and differences within the dataset, and allows for socially-relevant interpretations to inform policy development. [20]

Trustworthiness depends in part on the description of the analytical process. We first read all responses to become familiar with the data, then identified common and repeated themes to broadly classify the issues and topics of concern to respondents. This

preliminary content analysis was developed by two researchers (GGM, CLB) with qualitative research experience, who brought different lenses to the data set (one is male, early career, educated in the social sciences; the other female, health professional, and senior researcher). The preliminary topics and supporting evidence (data extractions) were discussed by all co-authors at a team meeting and draft findings developed, circulated by email, and additional comments and interpretations gathered through sequential iterations appraising data and interpretations. Because the patient partners were representatives of organizations each dedicated to different disease groups, their feedback served as a form of member-checking as to whether or not findings resonated with experiences and concerns of their respective groups. The four patient partners and four researchers thus co-constructed narratives reflecting the common experiences within the dataset and agreed upon quotes to represent each narrative. Collectively, the eight collaborators bring perspectives from men and women, young adult to late middle-aged, and health care, research, or lived experience across inflammatory skin, joint, and bowel diseases, experiences that contribute to the trustworthiness of interpretations. As a final step to enhance transparency and trustworthiness, the analytical process and findings were reviewed with a peer experienced in qualitative methodology and health research.

Results

We received 636 unique surveys. Respondents' age varied from 18-24 to 85-94 years (median age band 55-64 years) and 509 (80%) were women, which reflects the higher prevalence of women affected by most of the diseases in this study. The majority, 71%, were from British Columbia (the location of the newspaper with the survey link), with additional respondents from all other Canadian provinces and two territories. Most (91%) lived in a city

with at least one hospital. Forty-three percent reported multiple health conditions, often two of the three inflammatory disease categories part of PRECISION, e.g., Crohn's disease and arthritis. Consequently, the following proportions sum beyond 100%: 86% reported inflammatory joint diseases, 26% reported psoriasis, and 18% reported inflammatory bowel diseases.

Of the 636 respondents, 540 (85%) responded to the burdens and responsibilities question. These varied in length from a single phrase (e.g., "maintaining mobility and managing pain when I have flare-ups") to lengthy accounts of concerns for themselves and their families, descriptions of living with their disease(s) in daily life, and efforts take charge of their unique situation. Overall, responses outlined the ways in which the health care system and society in general are both helping and failing this population.

Four key narratives emerged from the large number of text entries: daily life disrupted; visible, invisible, and hidden disability; socio-economic vulnerability; and staying positive. Verbatim data show considerable overlap among the themes, therefore some quotes easily support more than one key narrative. Examples of social stigma, pain and fatigue, balancing work and family responsibilities, and worries about the future contributed to all four narratives. For example, *experiencing* symptoms like pain and fatigue were precursor to the first three narratives related to disruptions in daily life, disability perceptions, and social vulnerabilities, and *coping* with symptoms was apparent in staying positive, the fourth narrative. Each narrative is described below; alphanumeric labels link to quotations in Tables 1 to 3. Each quote references the sex, age band, province or territory of residence (using postal abbreviation), and reported diagnosis/es.

1. Daily Life Disrupted

Respondents told stories characterized by disruptions to tasks, activities, and roles, ranging from inconveniences to major shifts in how they participated in life. The most frequently cited antecedent to disrupted activities was persistent and sometimes unrelenting pain and fatigue, reported by more than half the sample. Managing symptoms necessitated setting priorities that tended to place obligatory work or household responsibilities ahead of equally important but more discretionary activities, such as maintaining social connections or enjoyable leisure activities (Table 1: A1, A2). Although employment was often stated as high priority, many respondents struggled to sustain participation in work. Repeatedly, respondents outlined difficulties fulfilling the roles that others expected of them, or shared serious concerns for the future if they were unable to continue work or take care of their own health (Table 1: A3, A4).

Descriptions of disrupted daily routines and the need for planning ahead were more often reported by those with joint or bowel diseases than those with skin conditions. Disruption was a prominent narrative in social situations and some found it very stigmatizing to “say no to social activities” and “curtail my hobbies and be vigilant of travel plans” in order to manage symptoms. Respondents experienced adversity in their social environments, feeling forced to adapt to circumstances and relationships that did not give credence to their illness experience (Table 1: A2). They reported concerns about being inadequate as friends, partners or family members, and some expressed feeling inferior to their peers at work. Respondents with inflammatory bowel diseases reported constant stress over whether or not they will be able to access a bathroom facility at a moment’s notice as curtailing social interaction (Table 1: B1, B2).

Table 1. Daily Life Disrupted.

Subtheme	Quotes
Accommodating symptoms disrupts work	A1: The largest burden is the effect fatigue has on social and work life - I have had to adapt my sleep patterns so I can perform at work; in the end my social life suffers. Female; 25-34; SK; rheumatoid arthritis (RA) A2: The disease is sometimes invisible and people don't understand issues with fatigue or sudden onset of pain/flares. This has a profound impact in the work place and in personal relationships. I am often perceived as lazy when I can't get mobile in the morning (late for work) and tend to over-compensate by working late and taking on more than I can manage. This pattern will lead to a flare which continues in a downward cycle. I hesitate to ask for help because I don't look sick (don't use a walker or cane) and am often judged to be 'weak.' In one workplace, a supervisor told my coworkers not to coddle me and that I had a low pain threshold. My partner has a hard time understanding the fatigue part but is very sympathetic to painful flares. Female; 45-54; BC; psoriatic arthritis (PsA) A3: I don't have the energy to do what I feel needs to be done, nor do I have the physical ability to do it. I want a pain free / tired free day. [I am] feeling inadequate in many aspects, losing my independence, always in pain, not being able to continue with my well-paying job. Female; 55-64; BC; RA, OA A4: Working full time and being a mom to 20 month old is very challenging, never mind finding time and energy to exercise, or even just be able to get enough rest. Female; 35-44; ON; RA
Disruptions specific to inflammatory bowel diseases	B1: The thing of most concern is that the only treatments mostly involving taking drugs with very significant and unpleasant (and dangerous) side effects. I know many with UC [ulcerative colitis] feel a bit like guinea pigs as we try to control our disease and be active and contributing members of society. Ulcerative colitis is very unpredictable. I have tried many "alternative" things, none of which have helped. I have experienced flares dozens of times and I have no idea why they happen when they do. The need for a washroom quickly is a huge burden. Also, public washrooms are neither private nor soundproof. It causes me emotional strain to wait in a stall until everyone has left, only to have someone else come it. It is embarrassing and very few people understand. I cannot always participate in activities with family and friends. I don't think they understand. I often feel they think I am making excuses (which I do sometimes to get around saying it is my colitis). I worry about the impact of my disease on my work. I worry very much that it will make my retirement less than what I dream of. I feel like a burden to my husband sometimes. I feel I complain too much although I try not to. I cannot talk to anyone about my actual symptoms as they are considered disgusting. Female; 55-64; BC; Ulcerative Colitis (UC) B2: Always looking to find a bathroom in case you need one, always having to explain to other people why you can't do or eat something, being disappointed in yourself because you can't do what everyone else is doing, disappointing your family (children) when you have to cancel plans at the last minute due to a flare up. Female; 65-74; BC; UC

Postal abbreviations: BC British Columbia, ON Ontario, SK Saskatchewan

2. Visible, Invisible, and Hiding Disability

Collectively, descriptions debated the extent to which these conditions are or are not visible, how that affects interpersonal relationships, and whether or not there is a need to consciously hide disability. A clear cluster of responses related to appearing sick versus well, of how "looking well does not always mean feeling well" and how this could be burdensome when trying to "give your family a break from your disease. Relationships take a beating." Visible disease characteristics, like psoriatic plaques, affected relationships (Table 2: C1, C2).

Although some respondents spoke to visible characteristics of their diseases, there were more descriptions of how invisible disability (appearing normal) led to individuals feeling marginalized (Table 2: D1) and wanting to explain, increase awareness, or find a way to foster understanding, assistance, or universal accessibility (Table 2: D2). Family, social and employment relationships were reported to suffer due to a lack of empathy and understanding: "*I guess the biggest casualty is that I never had the energy to create an active social life. A lot of people do not understand last minute cancellations for plans because all of a sudden you lack the energy to participate*" (Female, BC, 55-64, Psoriatic Arthritis). At times, respondents described feeling devalued by society as weak or dysfunctional, and how those attitudes are held or contested by their spouses, friends, coworkers or bosses (Table 2: D3, D4). In contrast, other descriptions related to the desire or perceived need to hide the disease. Sometimes this reflected wanting to participate in activities like anyone else, while other examples related to fears about being treated differently, or losing opportunities or jobs (Table 2: E1, E2).

Table 2: Visible, Invisible, and Hiding Disability

Subtheme	Quotes
Visible Disability	C1: Red raised patches and skin flakes all over my body interferes with my interpersonal relations and social life. Male; 55-64; AB; psoriasis C2: Unsightliness of plaques. Annoyance as I have psoriasis. People's comments and the unpleasant look of it. Female; 45-54; BC; psoriasis, Sjogren's syndrome
Invisible Disability	D1: Having an invisible disease comes with a lot of judgmental bigots' attention. Having accidents because of denied use of washrooms, being overweight because of the side effects of the medications. The pain can be crippling. Imagine not having ANY control over when and where you need to go to the washroom…while your body is in pain. Female; 25-34; BC; Crohn's, RA D2: [That] some people have a difficult time understanding or even believing I have to contend with illness can be trying. I was so surprised by what happened to me and by my diagnoses that I want to help others understand the complexities of rheumatic diseases. Female; 55-64; QC; ankylosing spondylitis (AS), psoriasis, Crohn's, Sjogren's syndrome D3: Chronic pain and unpredictable flare-ups make it hard to manage life on a daily basis. I always have to carry painkillers with me. Often, I have trouble riding a bus or subway because I have limited mobility and joint strength. Navigating in public is hard when others do not seem to understand that someone who looks 'normal' has trouble turning a doorknob, or holding a door open. Male; 55-64; BC; RA, PsA D4: Trying to manage a balance of work and family life while having chronic pain. Stress of not being able to support child if I have to take days off work. Being sick but not looking like a sick person is difficult as people don't understand. Female; 35-44; BC; lupus, Hashimoto's
Hiding Disability	E1: [It is a burden] making people aware of the disease without having a label put on you. Since my disease is not visible it is hard to hide pain. Female; 65-74; BC; AS E2: Being disciplined all the time. Not being a burden on my significant other and children. Hide the disease as much as possible from my employer. Male; 55-64; QC; AS, psoriasis

Postal abbreviations: AB Alberta, BC British Columbia, QC Quebec

3. Socio-Economic Vulnerabilities

Respondents explained how they simply didn't have the energy to concurrently maintain employment and family responsibilities and attend to their own health, which

resulted in financial strain (Table 3: F1, F2, G1). They spoke of “falling through the cracks” between healthcare and social systems because eligibility requirements for programs denied them access. They described experiences where health system or government priorities and budget constraints shifted definitions of disability in ways that excluded them from accessing the pensions or resources they needed or relied upon in the past (Table 3: G2, G3). Some respondents reported difficulty being taken seriously by their doctors (Table 3: K1, K3), and consequently suffered setbacks in their treatment and health, or expended time and effort coordinating and seeking out proper health care (Table 3: K2, K4). For those living alone, their living arrangement was frequently cited as exacerbating the negative effects that their disease(s) have on their quality of life (Table 3: F2, K4).

Repeatedly, respondents explained how their disease makes them economically vulnerable due to employment insecurity or loss and the high cost of treatment and medication. It was difficult to buy items like healthy food or services not funded by health plans to help them prevent complications (Table 3: H1, H2, H3). The high cost of biologics, as well as their unpredictable and potentially serious side-effects or worries about long term effectiveness, was a burden common to many respondents regardless of diagnosis (Table 3: J1, J2). The pressure and stress of dealing with health and social systems fostered a fear of the future and what it may hold (Table 3: J2).

Table 3: Socio-economic vulnerabilities

Subtheme	Quotes
Financial strain	F1: Full-time employment was not possible so there were financial concerns because I could only work part-time. Reduced finances because of part-time employment, coping with the physical challenges of household tasks like cleaning, shopping, cooking, having enough energy to socialize after doing essentials, not feeling like a burden to my family. Female; 55-64; BC; psoriasis, RA, OA F2: Chronic pain, immune symptoms, inflammation, quality of life. Financial

	burden of not working, being single and living alone in Vancouver. Having energy to eat well when I am flared up, and one of the biggest, is that patients really do need to be our own advocates in order to avoid falling through cracks in the system. Female; 35-44; BC; Endometriosis, inflammatory bowel disease
Disability programs are inadequate and restrictive	G1: Having a chronic, painful illness at a younger age means trying to juggle the symptoms (including crippling fatigue) with children, spouse, home responsibilities, and work. Combined with a long commute, it's almost impossible for me to spend any time taking care of myself, like getting more sleep or exercise, or eating better. I want to get better, and I know I need to take better care of myself, but I can't figure out how to make it work. I don't think there are any programs or support (at least I've never heard of any) for young people managing these types of issues. Most programs are geared towards the elderly. Female; 35-44; BC; RA G2: Trying to manage working, running a house and exercising on limited energy. Helping my children deal with the unknown and day to day issues of a chronically ill mom and finally, disability programs do not deal well with chronically ill people able to work part time Female; 45-54; AB; lupus, Sjogren's, polymyositis G3: How to earn a living. No money means no options. If the government keeps rejecting disability claims because chronic arthritis is not disabled enough how does one make money? Cannot go to swimming pool or buy decent quality food to help fight inflammation, cycle becomes a revolving door. Male; 35-44; BC; AS
Added costs of maintaining health with a chronic disease	H1: [I feel the] financial burden of not having a lot of (realistic) job options - costs associated with disease management that have no coverage (i.e., are not prescription drugs), "high cost, high quality foods," alternative health care, gym/pool memberships, equipment, living with chronic pain -not sleeping- not easy in social situations, (bathroom availability, food/water sources available). Female; 45-54; BC; UC H2 The cost of helpful therapies e.g., massage, medications, etc. [I'm] now on a disability pension, the things that support my quality of life are difficult to get. Female; 55-64; BC; lupus H3: I cannot afford to eat a good diet, and pay for my medications, treatments, supplements, and pay my bills on what I get from disability and the small amount I am allowed to earn. I have to choose to eat well or take the medications, I can't afford both. Without both, I cannot manage my disease well. Female; 35-44; BC; lupus
The cost of biologics	J1: My biggest concern is that the [biologic] which currently controls my RA could someday become less effective or stop working altogether, and that no other treatment will be effective. My other concern is that perhaps despite the [biologic], I could still be slowly incurring joint damage leading someday to disability and deformity. The greatest burden is managing the high cost of biologics. Female; 55-64; ON; RA

	J2: I'm fearful of what my future holds in terms of health problems. In addition, because of the symptoms I am unable to work or obtain employment and this impacts me severely financially. The medications I am on are also costly and again cause financial burden. Female; 45-54; BC; lupus
Time and effort to manage own healthcare	K1: My biggest concerns are: 1) Doctors listen to what I am saying about my symptoms and not rely solely on test outcomes. 2) Take seriously my description of being in severe pain. Female; 55-64; MB; Crohn's; OA K2: I have to be the captain (or co-captain) of my health care team. I have poor access to my medical records. There are a lot of out-of-pocket expenses. Important to be highly health literate, taking medications forever, social stigma, low level of socializing, hard to work full time, more disability in future. Female; 65-74; ON; RA, Sjogren's K3: Although I appreciate my doctors, I often feel they do not appreciate me asking questions or taking part in my own care. I am very experienced with this disease. I find they push for invasive diagnostic procedures promising no pain, when in the end there is pain. I know they are trying to help but I think treating UC [ulcerative colitis] patients is not their favourite thing. I wish health professionals learned more about the day to day challenges of living with a disease such as colitis. Female; 55-64; BC; UC K4: Constant pain, not knowing when my hip will pop out of its socket, long term disability from work (12+ years ago) and dealing with CPP [pension plan] and long-term shortage of money due to being unable to work and do the activities I would like to do, poor balance and living alone in a small island place when my present doctor and specialists are in the greater Vancouver area. I am NOT allowed to have 2 G.P.s - so must keep the M.D. who has helped me with my condition(s) for the last 12+ years and take the ferry and bus to appointments in Victoria and Vancouver as Travel Assistance Plan forms can't be forwarded to me as the Medical Clinic on this island won't give me TAP forms and the G.P. and staff in New Westminster have no idea how to locate these forms - a nightmare! Female; 55-64; BC; arthritis

Postal abbreviations: AB Alberta, BC British Columbia, MB Manitoba, ON Ontario

4. Staying positive

While the above three themes speak to undesirable consequences of inflammatory disease, there is a contrasting narrative arising from these written entries that tells a more positive story of resilience and adaptation. Examples of collaborative care, where health professionals and patients work together to ensure treatment both parties found appropriate, was one example. Some respondents shared strategies they found effective: *"I*

have received strong encouragement from my nephrologist and my rheumatologists to exercise, and a wonderful, now-retired rheumatologist referred me to a physio. Physical activity has been absolutely critical to my well-being. I'm so grateful!" (Female; 45-54; BC; gout, lupus, Sjogren's). Another respondent described collaborative care:

> *"My team of doctors listen to me, respect me and believe me. We work together to get me healthier. They are proactive in searching for answers and options. My acupuncturist and massage therapist have the same attitudes as the doctors. The very supportive environment this creates helps me stay focused on getting healthier; even when the pain is so bad I feel like I can't move, I know that it's my job to get out and walk and exercise. They're all doing what they can to help me and I, in turn, must do my part. Without their support, I'm sure there are many times I would have given up"* (Female; 55-64; BC; psoriasis, rheumatoid arthritis).

Staying positive is presented as a serious but necessary challenge. *"Pain management is my biggest burden. You must be constantly aware of your physical limitations and not make them your focus. Forcing yourself to stay active and positive despite how you feel"* (Female; 55-64; QC; ankylosing spondylitis). Keeping a positive attitude was cast by some participants as a struggle against fears and anxieties about the future. The following respondent shared reflections across several decades of living with inflammatory diseases, illustrating the episodic nature of both the disease and positive attitude:

> *"Initially (in my 20s) with total body coverage of psoriasis, I was concerned I would never meet anyone that could tolerate how 'ugly' I was (felt); then with onset of PsA [psoriatic arthritis] in my late 30's my main concern was keeping mobile and being*

able to care for our daughter. Now in my 70's after 10 years of clear skin and pain free joints due to the effectiveness of the [medication] injections, I'm concerned the drug is losing its effectiveness and I will lose the wonderful 'normal' life I have had – walking, cycling, dancing, sleeping, and pain free (almost) during the night" (Female, BC, 65-74, psoriatic arthritis, Sjogren's).

Discussion

Disruptions to daily life, systemic vulnerability, coping with (in)visible disability and staying positive are interconnected aspects of living with chronic inflammatory diseases affecting skin, joints, and bowel. Written passages support four intertwined narratives, none of which exists in isolation, illustrating challenges encountered on a regular basis, regardless of diagnosis. The reasons for disruptions differed across diseases and individual experiences, but the overall consequences were quite similar. For example, the difficulty of maintaining steady employment and income threatens financial stability; consequently one is less able to afford the goods and services that, alongside medical care, support a healthy lifestyle that makes the difference between inflammatory disease being a manageable condition rather than a miserable one. When daily life is disrupted, the relationships that hold peoples' lives together begin to unravel, whether it is a relationship to one's employer who sees inflammatory disease as a liability, or one's coworkers, friends or family who do not understand the burdens imposed by disease. Many survey respondents stated a need to try to hide their disability, having encountered or anticipated a lack of understanding or compassion from those around them, as essential to supporting a positive self-identity. Managing diseases, relationships, and life roles was a balancing act, consistent with prior

smaller, but more in-depth studies. [8 -13] Thus this survey of a large number of patients confirms the experiences described by others.

Some respondents regarded the responsibility to maintain a positive attitude while coping with chronic pain and disability as an ongoing mental and emotional challenge. Yet this was not a universal experience because other respondents appeared to have mastered a positive perspective. They dismissed disease-related challenges as part of life and focused on things that mattered to them, enjoying family, friends, and activities regardless of their health conditions. Because the survey item used the phrase “burdens and responsibilities,” it solicited responses regarding difficult experiences, yet the small number of respondents who spontaneously presented a positive narrative instead was nonetheless critical. What is unknown, given the limitations of a single, written submission from each participant, is the extent to which a positive perspective can be sustained by the individual’s resources such as access to health care, economic security, the presence of strong social networks, or responsibilities like caring for others – all of which contribute to health disparities. It is also possible that these descriptions of resilience, like inflammatory diseases, are episodic, or reflect a stage of adaptation to living with a long-term condition. [21] Based on the present findings, those with highly positive descriptions credited respectful, collaborative relationships with health care providers, and understanding family, friends, and employers with supporting their outlook on life.

The findings suggest that many respondents’ needs are not well-served by a system that isolates each individual problem to the exclusion of seeing the bigger picture. This bolsters evidence for a biopsychosocial approach that integrates the social experiences of patients with the psychological and physical impacts of their disease(s). Finding solutions to the consequences of long-term illness requires a patient-led research agenda because as

Rose argues, public/patient engagement are forms of civic participation and citizenship that work toward the democratization of science. [22] Patient engagement in research is an avenue for their concerns and priorities to be represented, and by extension, better addressed in health and social sectors. This confirmatory study with n=540 shows that many health needs are unmet from the patient perspective, explained in part by lack of attention to social determinants of health. That patients seek symptom relief, strategies to support daily life, a functioning social safety net, and empathic social support and health services is not new, but the repetition across multiple patient experiences indicates these important and long-standing issues have yet to be resolved.

Examples for engaging patients in research are widely available [23 -25] and our experience had both strengths and room for improvement. Researchers are generally motivated to try public engagement because they feel it will increase the relevance of their findings, while patients may be motivated by the desire for more user-oriented services. [23, 15] A moral rationale for patient-partnered research is that it honours and respects the patients' voice, supports participation, minimizes occupational disruption, and advances a role for patient organizations in public education of the need for societal supports, large and small. [24] Moving forward, a measure of patient engagement in research that can serve as a guide for assessing the quality and depth of patient engagement in a given project may be useful and lead to more user-oriented research. [25 -27]

Strengths and Limitations

The large sample in this study was a major strength as it ensured that all relevant topics to the study populations were uncovered. There are two key limitations. First, the survey was originally designed to inform research priorities and questions, and not as an original research study; thus, items were neither standardized nor pilot-tested. Second, the

single open-ended question is a minimalist form of data elicitation and while this paper presents a qualitative analysis it was not a prospectively designed qualitative study. Although the opportunity to probe further (as in other forms of qualitative inquiry) was not possible with this mode of collecting written narratives, we had narrative texts from over 500 Canadians. Typical qualitative research involves theoretically informed designs with in-depth descriptions from a small number of participants. What was lost in depth is counterbalanced by breadth, enhancing transferability to Canadians with similar diagnoses.

Qualitative studies of living with chronic inflammatory diseases have eloquently illustrated the burden and responsibility [3] [9] [13]; our survey extends those findings across a large number of people. The survey format allowed respondents anonymity and freedom to speak their thoughts, in contrast to the more personal interaction of a research interview. An advantage of this approach may be less likelihood of social desirability shaping responses, i.e., that respondents tell the researcher what they believe the researcher wants or expects to hear. The limitation, however, of having to take responses at face value without more probing means that some clarity of meaning may be lost. As a survey administered “by patients for patients,” a platform was provided for critical input from respondents that may otherwise be elusive in more structured quantitative and qualitative studies alike.

Public engagement in research happens most often at the stage when researchers need patient input to help identify a relevant research question. [28] While this was the case with our study, patient partners remained engaged throughout the research process, beyond the initial phase when it is advantageous to securing funding. We consider it a strength of this analysis that it was undertaken with respect to the values of patient and public engagement outlined by Gradinger et al., namely a concern for the ethical, political and

normative values, as well as for the process-based values such as respect, partnership and equality. [29]

Conclusion

Analysis of written responses to a survey created by patients for patients living with chronic inflammatory diseases shows many common experiences regardless of diagnosis, including disruptions to daily life and socio-economic vulnerabilities that create and contribute to worries about the future. Yet respondents also describe examples of patient-provider partnerships and social systems that contribute to personal resilience and capacity to participate in life. This paper illustrates a meaningful collaboration between patients and researchers that suggests a patient-led research agenda in chronic inflammatory diseases would foreground the role of the social determinants of health in shaping disease outcomes. Such findings should inform policy and service delivery through system change.

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