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Patients' views on the causes of their polymyalgia rheumatica: a content analysis of data from the PMR Cohort Study

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ABSTRACT

Objective: To explore primary care polymyalgia rheumatica (PMR) patient beliefs about the causes of their PMR.

Design: Qualitative content analysis was conducted on patients' written responses to the question of what they thought had caused their PMR. All data were coded and emergent categories of causal beliefs identified.

Setting: Community patients receiving primary care at general practitioner (GP) practices across England.

Participants: Participants were recruited from a primary care PMR inception cohort (n=654). Between June 2012 and June 2014 GPs referred 739 people with a new PMR diagnosis in the past 3 years into the study. Patients were mailed a baseline self-completion questionnaire, which included the question, 'What do you think caused your PMR?'. Responses to this question form the data set for the present study.

Results: 296 (45%) patients gave a possible cause for their PMR, while 276 (42%) respondents wrote 'no idea'. Common attributions include ageing (45, 18%), medication (18, 5%) and personal stress (53, 14%). 24 respondents (6%) thought their PMR was as a result of another medical condition.

Conclusions: This is the first study to examine causation beliefs in PMR, identifying a number of possible causes such as ageing, stress and as a complication of other medical problems. Understanding these patient beliefs may impact on treatment adherence and patient outcome.

Strengths and limitations of this study

- A strength of this study is the large sample size which is representative of the primary care polymyalgia rheumatica (PMR) population.
- Respondents in this study had been very recently diagnosed with PMR prior to questionnaire completion, therefore minimising recall bias.
- The content analysis approach used allowed for an unobtrusive and non-reactive way to study patients' opinions.
- Analysis of short written responses did not allow for consideration of broader contextual factors related to patients' beliefs, and therefore future in-depth qualitative research could give further insights.

suggest genetic factors including human leucocyte antigen shared epitope and polymorphisms in proinflammatory cytokines such as tumour necrosis factor-α and interleukin-6 cluster genes may be implicated.[2] Infections and vaccinations (eg, influenza) have also been linked to slightly higher risk of developing PMR, although to date, these studies are inconclusive.[2–6]

Patient beliefs about the causation of illness can have a significant effect on the attitude patients have about their illness, the stigma they attach to it and may impact on their health-seeking behaviour and attitudes towards treatment.[7] Qualitative studies of patients with rheumatoid arthritis (RA) suggest that patients believed their RA was triggered by a psychological factor or life event, whereas patients with spondyloarthritis (SpA) felt that their disease was as a result of a genetic predisposition.[8] These studies have helped clinicians gain better understanding of the views of patients with RA and SpA about the origins of their condition which may lead to improvements in care by promoting dialogue on the psychological impact of

INTRODUCTION

Polymyalgia rheumatica (PMR) is a common inflammatory disease of older adults, which is more prevalent in females than males and is predominantly found in the Caucasian community.[1] PMR classically presents with a sudden onset of bilateral myalgia/pain in the shoulder and hip girdles as well as systemic features such as weight loss, nausea and fever.[2] The aetiopathology of PMR is currently not well understood,[2] although studies

disease, and advice on suitable coping mechanisms and understanding expectations of treatment.[8] To date, there are limited data on patients' beliefs about PMR or its cause. This study aims to explore primary care PMR patient beliefs about the causes of their PMR.

METHODS
Patient cohort
Respondents in this study were recruited from a primary care PMR inception cohort (see ref. 9). Briefly, patients were recruited from 382 general practitioner (GP) practices across England. Between June 2012 and June 2014 GPs referred 739 people with a new PMR diagnosis in the past 3 years into the study. PMR diagnosis was made by GPs, who were provided with British Society for Rheumatology (BSR) guidelines on diagnosing PMR. Patients were mailed a baseline self-completion questionnaire. This included questions relating to PMR symptoms and severity, treatment, functionality, general health, lifestyle habits and sociodemographic factors, and included an open question, 'What do you think caused your PMR?'. Responses from this question provided the data for this study. All patients provided written informed consent.

Data analysis
All of the patients' written responses to the question about the causes of their PMR were first separated from the original questionnaires and stored on a separate password-protected spreadsheet for analysis, in order to ensure that written responses could not be linked to information in the questionnaires that could identify patients. The responses were then fully anonymised, with any potentially identifiable information, for example, names of people or places, removed or replaced by pseudonyms.

Respondents' answers were analysed using content analysis. This is a systematic method of ascribing meaning to qualitative data in order to identify recurrent patterns.[9] First, data were read repeatedly by a single author (MT) to achieve data immersion. Second, data were classified according to emergent descriptive codes that were identified. For example, the following response was coded as 'occupational strain': "…my work, climbing poles and ladders, leaning out on my belt connecting wires and services", while "…a road traffic accident as a backseat passenger in a stationary car outside my home" was coded as 'injury'. The following response was coded as 'no idea': "I simply have no idea".

Throughout the coding process, keywords were identified within the respondents' answers. This sometimes entailed picking up the main idea expressed in the sentence and making inferences as to what was being implied. For example, a response such as, "I don't know but my mum had it", indicates a hereditary cause, despite the participant not saying this explicitly. In these cases coding was more interpretive, moving beyond a face value description of the respondents' answers. An iterative, reflexive approach was adopted, whereby codes were continuously revised and refined throughout the analysis until data saturation was reached, with no new codes emerging.

The mean age and SD for the whole cohort was calculated. The number of participant responses within each preliminary code was counted. Codes that were seen to be closely related in meaning were then combined into broader categories, resulting in the identification of 11 main categories. The emergent categories were examined in relation to respondents' demographic characteristics.

A random sample of 100 responses was independently coded by another of the authors (BS) to check for intercoder reliability. Coders brought different disciplinary perspectives to the data (MT is from a clinical background, BS from a social science background), and the aim of independent coding was therefore to understand cross-disciplinary perspectives on the data and, through discussion, to come to an agreement on shared meanings and interpretations. The level of agreement between coders was therefore assessed through detailed discussion, rather than statistical calculation of levels of agreement.

The respective backgrounds of the two coders must be taken into account when interpreting the data, as their own subjectivities will inevitably have an influence on how the patients' responses are interpreted. However, this need not represent a limitation and can instead be seen to form an integral part of the analysis process. Despite their differing disciplinary perspectives, the two coders were largely found to agree on the main categories, and where any disagreement was apparent this was resolved through discussion of individual cases, which resulted in agreement on the final categories. This intercoder agreement can be seen to enhance the trustworthiness of the findings presented.

RESULTS
Main findings
Of those mailed, 654 (88.5%) returned the baseline questionnaire. Of the 654 responders 408 (62.3%) were female and the mean (SD) age was 72.4 (9.2) years. The majority of the sample were retired (n=513, 79.9%), with only 77 (11.9%) reporting themselves to be employed. The sample almost all identified themselves as being of white ethnicity (n=640, 98.2%). In terms of occupation, approximately a third of the sample had or previously held higher managerial, administrative or professional occupations, while 28% had intermediate occupations (eg, higher grade white collar workers) and 40% had routine occupations (semiskilled and unskilled work, eg, production, technical or operative job roles). Of the 654 respondents, 296 (45%) gave a possible cause in answer to the question, 'What do you think caused your PMR?'. Two hundred and seventy-six (42%) respondents wrote

'no idea' and 82 (13%) left the question blank. The 296 respondents who gave a possible cause were slightly younger, mean age of 71.4 (SD 9.9) years and 63% were female. The non-responders/'no idea' group had a mean age of 73.0 (SD 8.5) years and 62% of this group were female. Of those who suggested possible causes, 159 (54%) respondents reported multiple factors. The multiple factors given by respondents resulted in the total number of responses given (n=379) being more than the number of individuals (n=296). The initial number of codes totalled 37; though some related to answers provided by only one or two respondents. Eleven broader categories were identified from this initial set of codes. Some causes did not fall under a main category and therefore stood alone; for example, falls, muscle strain, road traffic accident injuries were all grouped to make one umbrella category of 'injury'; while 'personal stress', not falling under any category and having a large number of respondents, stood alone as a main category.

The main categories of PMR causes given by respondents were: occupational strain, personal stress, age and age-related conditions, medication, extension of an existing musculoskeletal disease, linked to another non-musculoskeletal illness, injury, genetic causes, environmental, lifestyle and lifestyle-related conditions, medical intervention complications and infection (table 1).

The three commonest causes cited were injury, personal stress, age and age-related conditions, respectively. The least common causes were environmental, consequence/complication of medical intervention and lifestyle and lifestyle-related conditions.

Causal attributions and participant variables

Respondents who attributed their PMR to age/normal wear and tear of the body had an average age of 74.5 years (range 58–91 years). This group was predominantly male (57% vs 43%) and mostly gave single causes for their PMR, for example: "My age, worn out joints" (79-year-old woman); "I would suggest age is the major factor" (78-year-old man).

Respondents who suggested that their PMR was a result of an existing musculoskeletal condition had an average age of 70.4 years (range 48–94 years) and were predominantly female (76% vs 24%). Only 21% of the individuals citing an existing musculoskeletal condition gave multiple causes. Several musculoskeletal diseases were mentioned, including the following examples: "I thought suffering for years with arthritis was the cause of PMR" (81-year-old woman); "I have no idea, although I do have Arthritis in knees, hands+think perhaps this may have been the cause" (76-year-old woman).

For respondents who attributed their PMR to personal stress, the average age was 67.2 years, and the age range was 38–89 years. These respondents were predominantly female (80% vs 20%). The stressful situations that were highlighted included bereavement, caring for a

terminally ill relative, stressful jobs and dealing with their own serious illness such as cancer or surgery. Thirty-eight per cent (20) of these individuals gave more than one cause. Examples include the following response: "Could be stress of change of lifestyle i.e.

Table 1 Participant reported causes of PMR and breakdown of the main categories (the main categories and their overall proportion relative to the sample as a whole given in bold)

Category	N (%)
Injury total	**63 (17)**
Falls	22 (35)
Leisure activities	14 (20)
Muscle and injury strain	18 (29)
Sports strain	9 (14)
Personal stress	**53 (14)**
Age and age-related conditions total	**45 (12)**
Wear and tear/ageing	43 (95)
Menopause	2 (5)
Related to other musculoskeletal diseases total	**40 (11)**
Arthritis (unspecified)	10 (31.25)
PMR relapse	8 (20)
Osteoarthritis	7 (22)
Rheumatoid arthritis	5 (16)
Fibromyalgia	3 (9)
Others	7 (16)
Infection total	**39 (10)**
Viral	22 (56)
Unspecified	10 (26)
Bacterial	2 (5)
Others	5 (13)
Occupational strain	**36 (9)**
Related to existing chronic diseases total	**24 (6)**
Type 2 diabetes	4 (12.5)
Anaemia	3 (9)
Thyroid problems	3 (9)
Weak immune system	3 (9)
Autoimmune disease	3 (9)
Others	9 (28)
Genetic	**21 (6)**
Medication total	**18 (5)**
Statins	13 (72)
Medication (general)	4 (22)
Polypharmacy	1 (5)
Consequence/complication of medical intervention total	**16 (4)**
Cancer treatment	7 (44)
Postjoint replacement surgery	5 (31)
Postsurgical complication	4 (25)
Environmental total	**14 (4)**
Harsh weather	12 (86)
Bad housing conditions	2 (14)
Lifestyle and lifestyle-related conditions total	**10 (3)**
Lack of physical activity	4 (40)
Diet	3 (39)
Excessive drinking and/or smoking	2 (20)
Obesity	1 (10)

marriage breakdown, new job, more responsibilities" (58-year-old woman).

With regard to infection, some respondents mentioned a history of travel infection with the inflammation and pain beginning subsequently. Others mentioned that they had an influenza vaccination before the PMR pain started; for example: "…chest infection, got admitted to hospital…" (73-year-old woman); "…flu jab…" (74-year-old man).

Another common cause was occupational strain. Manual occupations were specifically mentioned; including, delivery work, gardening, building and kitchen fitting jobs. Twenty-nine per cent of the respondents gave multiple causes and the most common of these was harsh weather (cold and damp) experienced while doing these jobs. This is not to say that the two were always connected, but 29% of people from this category mentioned both; as shown in the following examples: "Working in all-weather situations in 50 years as an electrician before health and safety rules" (73-year-old man); "…my work, climbing poles and ladders, leaning out on my belt connecting wires and services" (58-year-old man).

Poor lifestyle choices, including heavy drinking, smoking and poor diet were identified as potential causes: "Manufactured foods. Palm oil. White potatoes. Lack of Vit E. Lack of exercise" (73-year-old woman); "For certain—neglecting my health through bad habits over 15 years—i.e. drinking/smoking too much, not eating regularly…" (67-year-old woman).

Statins were the commonest group of drugs to which respondents attributed PMR, accounting for 72% (13) of those that attributed their PMR to medication: "After my heart attack on 28th Dec 2013 my prescription for Atorvastatin was increased from 20mg to 80mg. 7 weeks later my PMR symptoms started. I was taken off Atorvastatin and later back on. My PMR became worse" (84-year-old woman).

Respondents also said their PMR resulted from medical intervention, in particular surgery, with some highlighting a knee or hip replacement that had put extra strain on their shoulders or the other leg, which they felt contributed to their PMR.

Genetic causes were mentioned by 9% of the cohort. The mean age of this group was 65.8 years and 75% were female. This factor was largely given as a single cause; except for 15% of the respondents who coupled genetic causes with occupational strain, personal stress and a viral infection.

DISCUSSION

This study has identified a wide variety of factors that respondents believe caused their PMR. Commonest proposed causes were injury, personal stress and the ageing process. While some respondents cited a single cause; others suggested multiple factors in the development of their condition. However, the majority of those in this cohort did not know what had caused their PMR. This study is the first to examine patient beliefs about the causes of PMR.

One strength of this study is the analytical framework used. Using a summative content analysis approach was advantageous because it is an unobtrusive and non-reactive way to study opinions.[10] Additionally, independent coding by two researchers, who were able to achieve a high level of agreement in their coding of the data and subsequent detailed discussions, improves the credibility of the results. A further strength is the large sample size which was composed providing a representative sample of the primary care PMR population. Furthermore, respondents in this study had been very recently diagnosed with PMR prior to questionnaire completion, minimising recall bias. A limitation of the study, however, is that an analysis of short written responses does not allow for consideration of broader contextual factors related to patients' beliefs. For instance, patients were not asked about the information sources they had drawn on in forming these causal beliefs. We therefore do not know how many patients had actively investigated causation themselves, either online or in discussions with treating clinicians, or whether patients were basing these beliefs solely on experiential and/or anecdotal evidence. Future research could therefore build on the breadth of views accessed in this study through an in-depth qualitative investigation of people's perceptions, allowing potential causal explanations and their underpinnings to be explored in greater detail.

A varied range of causal beliefs have also been observed in relation to patients with arthritis[11] and similar studies of patients with arthritis have shown that opinions about causation influence the psychological aspect of the disease, which may in turn affect coping.[8] This may also be the case with PMR because of the risk of serious complications such as giant cell arteritis, which untreated can lead to blindness.[12] A study on Latina women with arthritis stressed the need for having emotional and social support to deal with these psychological aspects of rheumatological disease,[13] and as such understanding illness beliefs may help improve coping for patients with PMR.

A study of the impact of illness beliefs in arthritis (of various forms, including osteoarthritis and RA) on patients in the Dominican Republic showed that patients believed that their arthritis was caused either by contact with water or that it was a test from God. As a result, patients avoided contact with water and prayed as a way of coping. Although these views are unconventional, the study showed that having these passive explanatory models[14] help patients accept their illnesses.

A sizable proportion of respondents in this study said that their PMR was caused by the normal ageing process. This tendency to normalise illness has also been shown to be common in patients with arthritis;[15 16] Sanders et al,[15] for instance, reported that their interview participants, aged 51–91 years, 'referred repeatedly to a natural degenerative theory of causation' for their arthritis. While such an explanation can help in the acceptance of the illness, it may also lead to a delay in seeking care. Other common causes cited, such as stress and injury, show similarity with

the findings of Berenbaum et al[8] who explored patient beliefs in France about RA and SpA through in-depth interviews. In relation to RA, they found that personal stress, often resulting from a distressing life event, was frequently cited as a trigger for onset of the condition, as was intense physical activity, which may be comparable to the category of 'injury' identified in the present study, found here to include 'muscle and injury strain' and 'sports strain'. The similarities with Berenbaum et al's[8] findings suggest that causal beliefs about PMR are unlikely to be entirely unique to the condition, and may instead share similarities with beliefs relating to other rheumatic conditions, particularly RA, perhaps reflecting the inflammatory nature of both conditions.

While citing the natural ageing process as a cause positions the onset of PMR as somewhat inevitable and therefore out of the patient's control, beliefs about personal stress and injury suggest external factors—both psychological and physical—that patients may have some control over; perhaps suggesting a degree of agency on their part. The degree of agency patients perceived themselves as having over their PMR could have implications for how they view their ability to self-manage the condition; therefore, understanding these beliefs can also have implications for the clinical advice given to patients about self-management of PMR.

Some of the causes of PMR that were cited are medically unlikely; for example, diet and insect bites. While it can be unhelpful if patient beliefs are at odds with clinical knowledge, understanding these beliefs enables them to be addressed and may help patients make sense of their condition. Therefore, even if patients' beliefs are unlikely to reflect reality, they can be important for how patients cope with their condition, and therefore these beliefs are no less valid. For example, in the case of the patients in the Dominican Republic, although their belief that water caused their arthritis was not correct, it was because of this belief that they attached great importance to draining knee effusions, and this lead to care seeking and improved disease management.[14]

Finally, a study by Lambert et al,[17] regarding patient and physician views on arthritis suggested that exploring patient beliefs about disease causation can help in identifying and addressing unhelpful beliefs. For example, if a patient expects that the medication will eliminate pain and reverse joint deformity, when this outcome is not achieved patients may struggle to trust their physicians.[17] Exploring patients' beliefs of causation, prognosis and treatment helps in allaying fears, patient education and treatment adherence and can result in more realistic patient expectations.

The findings presented in this study suggest a need for greater dialogue between patients and clinicians, so that clinicians explore such thoughts/ideas in order to dispel any unhelpful illness beliefs.[8] For example, if a patient harbours the belief that exercise may have caused their PMR, this may deter them from even doing the light joint exercises that are recommended as part of PMR management. While addressing such beliefs should already be part of routine clinical practice, making clinicians more aware of the most common causal beliefs held by patients with PMR may place them in a stronger position to identify and address unhelpful patient beliefs, particularly in cases where patients are not forthcoming in expressing these beliefs in the consultation. The findings presented here can therefore provide clinicians with a basis from which to explore these beliefs in relation to individual patients.

This study provides interesting patient insights into the cause of their PMR and may prompt additional hypotheses that warrant further study. Some patient attributions including infection and age have been postulated previously,[2–4] although no definite evidence exists at present. Likewise no clear evidence exists at present regarding an association between either injury or stress and PMR onset; however, though not conclusive, some studies have found an association between stress and increased disease activity in certain rheumatic conditions,[18] and it may therefore be useful to explore this association in future research specifically in relation to PMR. Some patients attributed their PMR to medications, especially statins. Myalgia is a commonly reported side-effect of statins, and this finding lends support to evidence from de Jong et al[19] who found an association between statin use and the occurrence of PMR. However, to date there is no evidence of a causal relationship between statins and PMR, and therefore further research exploring the nature of this association would be beneficial.

Thus, to date, although the cause of PMR is not known, patients with PMR reported a range of factors which they believe to have caused their illness. Commonly cited causes included injury, personal stress, ageing, medication and chronic illness. Engaging in dialogue with patients to understand their illness beliefs may help patients to make sense of their illness and influence coping and self-management strategies and thus have a positive impact on the clinical management of PMR.

Competing interests None declared.

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