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Idiopathic Pelvic Girdle Pain as it Relates to the Sacroiliac Joint

Building a Collaborative Model of Sacroiliac Joint Dysfunction and Pelvic Girdle Pain to Understand the Diverse Perspectives of Experts

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Abstract

Background: Pelvic girdle pain (PGP) and sacroiliac joint (SIJ) dysfunction/pain are considered frequent contributors to low back pain (LBP). Like other persistent pain conditions, PGP is increasingly recognized as a multifactorial problem involving biological, psychological, and social factors. Perspectives differ between experts and a diversity of treatments (with variable degrees of evidence) have been utilized.
Objective: To develop a collaborative model of PGP that represents the collective view of a group of experts. Specific goals were to analyze structure and composition of conceptual models contributed by participants, to aggregate them into a metamodel, to analyze the metamodel's composition, and to consider predicted efficacy of treatments.
Design: To develop a collaborative model of PGP, models were generated by invited individuals to represent their understanding of PGP using fuzzy cognitive mapping (FCM). FCMs involved proposal of *components* related to causes, outcomes, and treatments for pain, disability, and quality of life, and their *connections*. *Components* were classified into thematic *categories*. Weighting of *connections* was summed for *components* to judge their relative importance. FCMs were aggregated into a metamodel for analysis of the collective opinion it represented and to evaluate expected efficacy of treatments.
Results: From 21 potential contributors, 14 (67%) agreed to participate (representing six disciplines and seven countries). Participants' models included a mean (SD) of 22 (5) *components* each. FCMs were refined to combine similar terms, leaving 89 *components* in 10 *categories*. Biomechanical factors were the most important in individual FCMs. The collective opinion from the metamodel predicted greatest efficacy for injection, exercise therapy, and surgery for pain relief.
Conclusions: The collaborative model of PGP showed a bias toward biomechanical factors. Most efficacious treatments predicted by the model have modest to no evidence from clinical trials, suggesting a mismatch between opinion and evidence. The model enables integration and communication of the collection of opinions on PGP.

Introduction

Pain in the lumbopelvic region is an enormous issue globally and the leading cause of disability in the developed and developing world.[1] Despite the enormity of this problem, much remains to be learned about the underlying causes for the condition, and the most effective preventative and treatment strategies. A major issue

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is that in most cases the underlying cause/mechanism is unknown.[2] Many different structures may be involved[2–4] and pain may be maintained by central sensitization rather than by ongoing nociceptive input from the periphery.[5] Of the many potential sources of nociceptive input that may contribute to the pain experience, one structure that has been particularly controversial is the sacroiliac joint (SIJ). Although some argue that dysfunction of the SIJ is a common contributor to low back pain (LBP) (45% of individuals with chronic LBP below L5),[6] others consider it to have infrequent involvement (10%-13% of individuals with chronic LBP).[7–9]

Dysfunction and pain associated with the SIJ has been suggested to have specific characteristics (eg, pain location and provoking activities)[9–11] and is considered to be prevalent in conditions such as pelvic girdle pain (PGP: defined as pain experienced between the posterior iliac crest and the gluteal fold, particularly in the vicinity of the SIJ), often in association with pregnancy.[10] Differential diagnosis for other causes of LBP has been based on responses to specific pain provocation[11] and movement tests.[12] Multiple disorders and mechanical dysfunctions have been proposed. From a movement perspective, dysfunctions such as joint laxity,[13] failed load transfer[14] and abnormalities of joint alignments[15] have been proposed, with variable evidence.[16,17] The SIJ can also be a characteristic site for specific rheumatological conditions (eg, ankylosing spondylitis).[18] Because of the diversity of proposed mechanisms, there is parallel diversity of treatments offered, again with inconsistent evidence.[2,10,19] Current literature lacks consensus regarding mechanisms, contributing factors, and treatments, and vastly different views are held by different professional groups. It was the contention of our study group that advances could be made in our understanding of PGP (the term selected in this study to include SIJ dysfunction/pain) by building a model that included the diversity of conceptualizations of the condition.

Collaborative modeling is a participatory method that aims to gather the diverse opinions of individuals to build a single model inclusive of all ideas that delineate the scope of a problem.[20] Fuzzy cognitive mapping (FCM) is a collaborative modelling technique that elicits participants' mental models about a problem through a networked structure of concepts and their causal interdependency.[21] The term "fuzzy" relates to the fact that each connection was given a weighting, based on expert's opinion, to indicate the strength and direction of effect.

This approach incorporates a wide range of conceptualizations into a standardized format that can be used to illustrate and interpret the problem, and even to simulate possible solutions.[20] Although the main objective of collaborative modeling is to synthesize and share knowledge, the analysis of the structure, composition, and functionality of FCM models enables identification of core assumptions, evaluation of the relative importance placed on different concepts and testing of various scenarios, such as the impact of a treatment. This approach has recently been used to build a collaborative model of LBP using opinions of a broad range of experts.[22] The resultant model has highlighted that despite various disciplinary backgrounds of contributors, psychological features are considered to have the strongest importance in LBP.

The overall objective was to develop a collaborative model of PGP/SIJ dysfunction with contribution of experts across a diverse range of disciplines. The specific aims of this study were to analyze the structure and composition of the models generated by individual contributors, to aggregate them into a metamodel, to analyze the composition of the metamodel, and to use the metamodel to identify the group's overall estimation of the relative efficacy of treatments when all contributors' opinions are combined.

Methods

To build a collaborative model, 14 individuals with research and clinical expertise in PGP (purposefully drawn from different disciplines), generated individual FCMs that incorporated all of the *components* that they considered to be relevant for the problem (eg, causes, consequences, and treatments) and the *connections* between them.

FCMs are semi-quantitative models that allow for the analyses of the composition, structure, and behavior.[23] The composition of the FCMs includes the qualitative concepts participants used to characterize the problem. Accordingly, researchers frequently suggest the comparison of composition of the models to quantify similarities or differences of contents.[24,25] Moreover, examining the structural characteristics of FCMs demonstrates how people view the interconnectedness of system components through a network of nodes and connections. These analyses aim to obtain valuable information from the structure of the maps through a set of network metrics (eg, number of nodes, number of connections, centrality of concepts, density, and complexity).[21,26] These metrics can be interpreted as indices for comparing the structure of FCMs and therefore highlighting the cognitive diversity of participants.

FCM models also enable the quantitative assessment of the behavior of the system using simulations.[27] Scenarios can be run using FCM computation that enable simulation of impact of a particular input (eg, the impact of a treatment or change in a risk factor). The differences in key elements of the system when specific aspects of the model are changed are represented in the results of scenario analyses[23,26,28,29] which characterize how participants perceive the behavior of the system.

Potential participants were identified by members of the investigative team (P.H., J.C., J.P.) through extensive search of the literature and speaker's lists of relevant conferences, and through discussion with other experts in PGP.

Potential contributors were considered eligible for inclusion if they represented major disciplines in research/management of PGP and there was evidence that they had made a substantial and ongoing contribution to the literature related to PGP, as evidenced by at least two of the following: (1) Contribution to at least three published works in the preceding 3 years; (2) Keynote/invited presentations at major meetings related to LBP/PGP; (3) Contribution to major working groups/committees of LBP organizations; (4) Contribution to organization of major LBP/PGP meetings/conferences; (5) Contribution to LBP/PGP texts; and (6) Contribution to clinical practice guidelines/systematic reviews. From a total of 21 invited contributors, 14 (67%) agreed to participate (Table 1). The study was granted exemption from the Michigan State University Institutional Review Board.

The individual FCMs were built during a 1-1.5-hour semi-structured interview using videoconferencing and the freely available *Mental Modeler* software developed by SG.[30] Each participant was initially presented with three *components*: - "Pain," "Disability," and "Quality of Life," representing main outcomes of living with pain associated with PGP. Participants were then asked to name additional *components* (major factors contributing to PGP) that they considered would "directly affect" these three outcome *components*, and to consider all possible interactions between *components* (including feedback loops) in their model. As a new *component* was added, the participant was required to confirm the direction of the relationship, whether it caused an increase or decrease in the *components* it was connected to, and the strength of each *connection* between −1 and 1. After completion of the inclusion of *components*, participants were asked to identify the treatments that they considered would impact directly or indirectly the three main outcomes of PGP, identify pathways for this impact (*connections*), and to nominate the strength of these *connections*. Sessions were recorded with the consent of the participants for later clarification of meaning of elements of the model.

Table 1
Disciplines and countries of participants (10 males and 4 females)

Discipline	Subdiscipline	Country	Number
Physical Therapy			5
	Clinical (3)	Canada; New Zealand; United States	
	Musculoskeletal research (2)	Norway; Sweden	
Orthopedic Surgery		Sweden; United States	3
Physical Medicine & Rehabilitation		The Netherlands; United States	3
Anatomy		The Netherlands	1
Osteopathy		Australia	1
Sports Medicine		Australia	1

The study core team (P.H., J.C., J.P., A.L.) reviewed the *components* present in the initial 14 FCMs and modified them into a standardized format either by using the terms selected by the participants or a term of synonymous meaning from a list of standardized terms. This was done to enable aggregation of FCMs contributed by individual experts. The standardized terms were defined based on outcomes from several phases of consultations and a consensus meeting with the participants in the similar study concerning LBP.[22] As a result of this process, the structure of the original FCMs (structural features such as the number of *components* and *connections*) or composition (the issues represented by the *components*) did not change, but the components with similar meaning in different FCMs were combined to form a smaller number of standardized unique terms (from the original 312 to 89). These terms were then allocated to 10 *categories* (Table 2). (For a more detailed description of the process of refining terms and *categories*, see reference 22.)

The structure and composition of FCMs were analyzed using the graph theory.[20] The following metrics were calculated (adopted from Cholewicki et al[22]).

FCM structure:

1. *Total Components (N)* - number of *components* included in an FCM
2. *Total Connections (C)* - total number of *connections* in either direction included in an FCM
3. *Density (D)* - number of *connections* as a proportion of the number of all possible *connections* in both directions (see Appendixs S1 for equation)
4. *Connections per Component* - average number of *connections* in either direction per *component*
5. *Complexity Score* - calculated as the ratio of Receiver/Driver *components* (total number of *components* that only have inputs/total number of *components* that only have outputs) and provides a measure of the degree to which effects of Drivers are considered.

FCM Composition:

1. *Sum of Centrality (Sc)* - centrality (c_i) measures the weighted contribution of each *component* within the FCM. Sc is then calculated as the sum of centralities of all *components* in a *category*. A standardized Sc (NSc) score was calculated by normalizing the Sc for each *category* to the total Sc for all *categories*, excluding "Outcomes" and "Treatment/ Intervention" for each FCM (see Appendix S1 for equation). *Components* in the FCM with the highest centrality values are considered the most important.
2. *Cognitive Color Spectrum* - color bar chart that demonstrates the sequence of dominance of *categories* in a participant's FCM. It is generated by sorting the NSc of each *category* by their color starting from the most central *category*.

Table 2
Categories for allocation of FCM *components*

Category	Definition
Behavioral/Lifestyle	Lifestyle "choices" including: smoking; sleep; physical activity; diet; insufficient time.
Biomechanical	Factors that determine/cause/relate to tissue loading including lifting; posture; motor control; muscle imbalance; etc.
Comorbidities	Conditions that are comorbid with PGP and pain such as: rheumatoid arthritis; cancer; or diabetes.
Individual	Factors that are part of the "make-up" of the person including: age; body weight; physical capacity; strength; genetics; and individual features thought to predispose to PGP and pain such as prior history.
Nociceptive detection and processing	Biological factors related to pain/nociception including: sensitization; neuroimmune interaction; "neuromatrix", etc.
Psychological	All aspects related to psychology including: fear of pain/(re)injury; catastrophizing; self-efficacy; etc.
Social/Work/Contextual	Factors related to work and relationships including: work support; family environment; social status; spirituality/religion. Includes factors that are external to the person such as environmental/policy, access to treatment; political, physical environmental, social, cultural context.
Tissue injury or pathology	Biological factors of tissue/systems including: tissue injury; disease; pathology; cytokines; and consequences/outcome of loading rather than the mechanisms that cause loading, which are categorized as "Biomechanical."
Outcomes	Core outcome measures included in every model were "Pain," "Disability," and "Quality of Life."
Treatment/Intervention	Any intervention for treatment and prevention of SIJ pain.

Adapted from Cholewicki et al.[22] ICF = International Classification of Functioning, Disability and Health.[61]

3. *Cognitive Diversity Index (CDI)* – quantitative measure that reflects how many different *categories* are represented in an FCM, and simultaneously considers how evenly the *components* are distributed among those *categories*. A higher value indicates that an FCM has *components* representing more *categories* and contributing more evenly to these *categories*, whereas a lower value indicates fewer *categories* and bias toward specific *categories* (see Appendix S1 for equation).

The individual FCMs were aggregated into a metamodel that represented the group's view. Because we did not have any data regarding the credibility of the concepts by which to weigh them during the aggregation, a simple FCM averaging method with zeros was used.[31,32]

In this method, each individual FCM was converted to the adjacency matrix and augmented to include all unique components present in all FCMs after the refinement of terms, resulting in the same matrix size 89 × 89 for all FCMs. The connections between *components* not mentioned in the original FCM created by a participant were given "zero" weights in his/her individual FCM, representing "dummy" concepts added to the model. Subsequently, the metamodel was constructed by averaging *connections* across the adjacency matrices:

$$\boldsymbol{a} = \frac{1}{M}\sum_{i=1}^{M} a_i,$$

where M is the number of participants, $\boldsymbol{a}$ is the connection matrix of the aggregated model, and $\boldsymbol{a_i}$ is the connection matrix of the model developed by *i*th participant (in this case, 14 participants).

The metamodel was used to study the relative emphasis placed by the group on various *categories* (eg, Psychological vs Biological factors) by computing *Sc* for each *category* and it was also used to evaluate the group's collective view of relative efficacy of different treatments by performing scenario simulations. The metamodel can receive an input concept by initializing one or more of its *components* to a value between 0 and 1. During simulation, metamodel state is iteratively calculated until it converges by propagating the initial values throughout the metamodel network according to the weights between its *components* and a threshold function.[33] The final values of the *components* representing the output concept are examined to assess the relative effect of various input concepts. To assess the relative efficacy of different treatments, they were individually initialized to 1 in each simulation and the resultant values of "Pain," "Disability," and "Quality of Life" outcome components were recorded. The simulations were performed using a custom-written software in Python (Python Software Foundation, www.python.org) with a sigmoid threshold function.[34]

Results

The individual FCMs ranged from 14 to 32 *components* that were linked by between 25 and 125 *connections* for an average of 2.2 (SD = 1.1) *connections* per *component* (Table 3). In general, density was inversely related to the number of *components*, that is, when more *components* were included, fewer of the total possible *connections* were made. This relationship reached statistical significance when one outlier (#9) with a large number of *connections* was omitted from the calculation ($R = -0.80$, $P = .001$). Examples of two models with substantially different complexity and density scores are presented in Figure 1.

Table 3
Metrics describing structure of FCMs for each participant

FCM no.	Total comp.	Total connections	Density	Connections per comp.	Complexity score	Cognitive diversity index
#1	20	46	0.121	2.3	0.111	6.21
#2	20	39	0.103	2.0	0.077	5.99
#3	26	51	0.078	2.0	0.053	5.65
#4	25	41	0.068	1.6	0.111	5.04
#5	21	37	0.088	1.8	0.077	4.88
#6	26	41	0.063	1.6	0.067	4.74
#7	16	28	0.117	1.8	0.111	4.62
#8	21	54	0.129	2.6	0.000	4.61
#9	23	128	0.253	5.6	0.000	3.96
#10	14	38	0.209	2.7	0.125	3.74
#11	18	25	0.082	1.4	0.083	3.72
#12	23	40	0.079	1.7	0.000	3.67
#13	27	49	0.070	1.8	0.067	3.63
#14	32	51	0.051	1.6	0.043	3.61
Mean	22	48	0.108	2.2	0.066	4.58
SD	5	25	0.057	1.1	0.043	0.90
Min.	14	25	0.051	1.4	0.000	3.61
Max.	32	128	0.253	5.6	0.125	6.21

Comp. = *component*; No. = number. Order of FCM numbers is identical to that used in figures.

The most frequent *category* with the highest Sc in the individual FCM models was "Biomechanics" (five models) followed by the "Social/Work/Contextual" *category* (three models) (Figure 2). When thematically related *categories* of "Biomechanics" with "Tissue injury or pathology" (biophysical factors) and "Psychology" with "Social/Work/Contextual" (psychosocial factors) were grouped, seven and four FCMs gave the highest Sc to the biophysical and psychosocial factors, respectively.

The metamodel, an aggregate of all individual FCMs, which reflected collective group thinking, consisted of 89 *components* and 372 *connections*, and is presented in Figure 3A (Interaction version final metamodel is available in Appendix S2). In this metamodel, the "Biomechanical" factors (Figure 3B) had the highest Sc, followed by "Psychological" (Figure 3C), and "Behavioral/Lifestyle" factors (Figures 2 and 4). The "Comorbidities" and "Nociceptive detection and processing" *categories* had the two lowest Sc scores in the metamodel. The centrality attributed to each *component* of the metamodel is presented in Table 4. These data show the relative weighting placed on each *component* within each *category* and overall. The *components* with greatest centrality overall (other than outcomes) were: Cognitive (3.123) (Psychological) (eg, expectations, beliefs and perceptions concerning pain); Good Physical Activity (1.724) (Behavioral/lifestyle); Poor Sleep (1.094) (Behavioral/lifestyle); Inflammation (1.079) (Tissue injury or pathology); Poor Anatomical/Structural characteristics (1.012) (Biomechanical); Motor Impairment (0.982) (Biomechanical); Poor Posture and Alignment (0.876) (Biomechanical); Employment (0.870) (Social/Work/Contextual factors); Access to Support Networks (0.838) (Social/Work/Contextual factors); and Tissue Damage (0.794) (Tissue injury or pathology).

Although none of the individual FCMs included *components* from all eight *categories* (half of the models included *components* from five or fewer *categories*), the metamodel *components* represented all *categories* (Figures 2, 4, and 5). Therefore, as expected, the CDI of the metamodel (6.39) was higher than CDIs of any of the individual FCMs (Figure 5).

The results from the metamodel simulation of various treatment interventions identified by the participants are presented in Figure 6 as the effects on "Pain," "Disability," and "Quality of Life" relative to the most effective treatment. These metamodel simulation results, which summarize the collective opinion of all contributors, suggest that the interventions expected to be most effective for reducing "Pain" are injection, exercise therapy, and SIJ surgery. Exercise therapy, cognitive behavioral therapy, and advice/education were considered the most effective interventions for reducing "Disability." Exercise therapy, cognitive behavioral therapy, and SIJ surgery were the interventions considered to be the most effective for improving "Quality of life." Acceptance therapy (psychological therapy that teaches mindfulness skills to deal with the uncontrollable experience of pain[35]) had the smallest expected effects on the three outcomes.

Discussion

This study produced a collaborative model of PGP that represents the collective view of the experts across a range of disciplines. The model shows how expert opinions differ between consideration of PGP and LBP, and how opinions relate to current evidence for treatments.

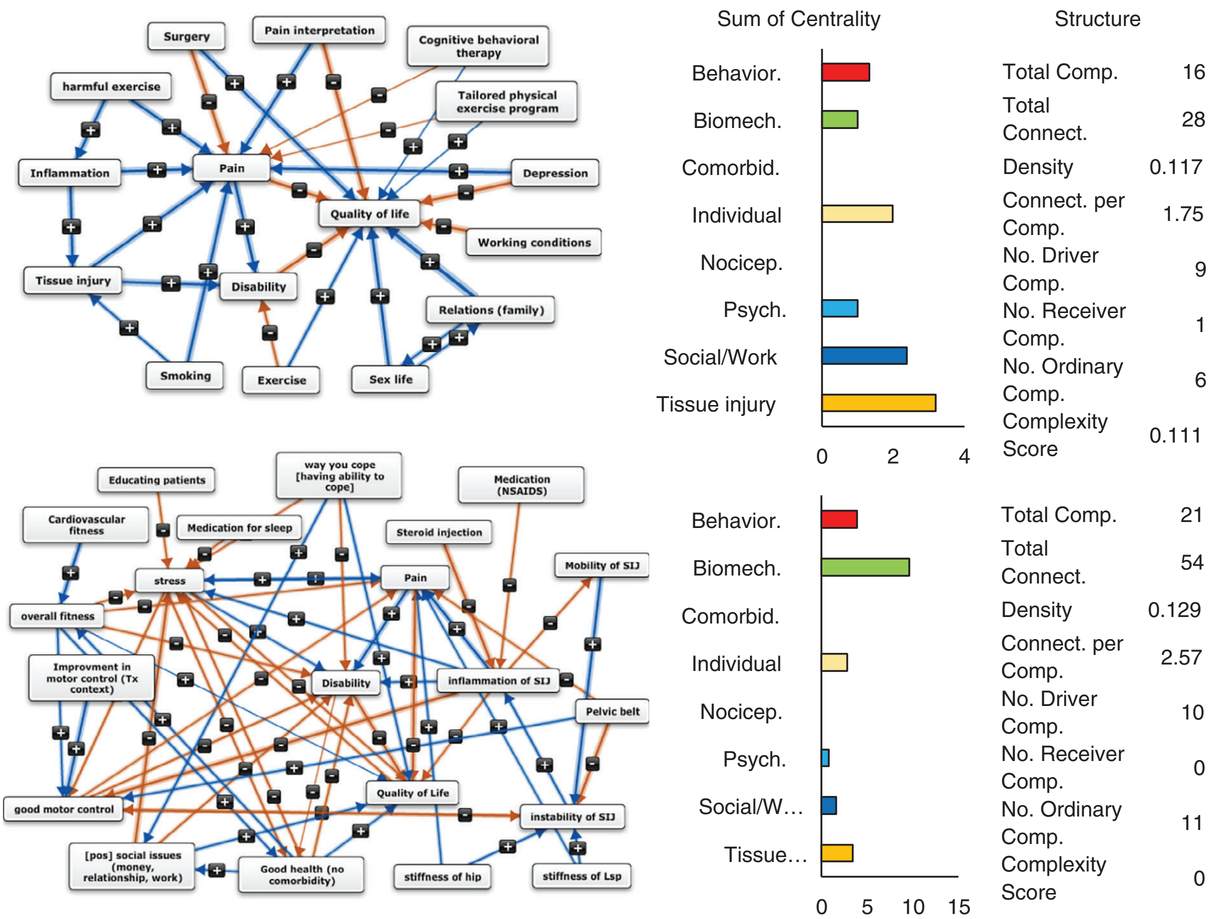

Figure 1. *Fuzzy Cognitive Maps (FCMs) for two representative participants with different structure and composition.* The FCMs that were generated by the participants are shown (left) in their original form, prior to refinement of the *component* terminology. The Sum of Centrality (Sc) (middle) and structural features are shown for the final FCM after refinement. Note the different Sc and range of structural features that characterize the models of different participants considering the problem of PGP and pain. Comp. = *component*; Connect. = *connection*. (See Table 3 for full titles of *categories*.)

Comparison of Structure and Composition of Individual Models of PGP and LBP

Individual FCMs for PGP were diverse in their structure with 14–32 *components* and 25–128 *connections*, but were, on average, less for FCMs of PGP than LBP (*components*: 22 vs 25; *connections*: 48 vs 77) with fewer connections per component (2.2 vs 3.1).[22] Although this implies that PGP was generally considered to be less complex than LBP, most of the same *categories* were considered to be relevant. A notable exception was that only one FCM for PGP included *components* related to "Nociceptive detection and processing" (eg, central sensitization) in comparison to the 18 of 29 FCMs for LBP. Limited recognition of this issue is at odds with the growing recognition of such processes in maintenance of pain,[36] including PGP[5] and appears consistent with the tendency toward biomechanical conceptualization of SIJ dysfunction (see below).

The *categories* with greatest centrality in individual FCMs differed from that reported for LBP. Whereas "Psychology" was the *category* with greatest centrality for nearly half of LBP FCMs (14/29), this was identified for only 1 of 14 FCMs for PGP. This concurs with the general view of the literature regarding PGP. For instance, the European Guidelines for the Diagnosis and Treatment of PGP state that "based on the present limited knowledge, the impression is that yellow flags (psychosocial features) are less common among PGP patients than among LBP patients."[10]

In contrast to LBP, the *category* that most frequently had the highest centrality in PGP FCMs was "Biomechanics" (5/14 compared with 4/29 for LBP), and if considered along with the related *category* of "Tissue injury or pathology," this accounted for 7/14 of the FCMs. The greater bias toward biomechanics and tissue injury may have several explanations. First, the term "PGP" attributes the condition to a specific anatomical structure, which contrasts the case for LBP. This could have led to the participants' interpretation of a more mechanical foundation and stronger attribution to tissue-level effects. Second, there has been considerable emphasis on biomechanical models of SIJ function and dysfunction,[13,14] which has strongly influenced both

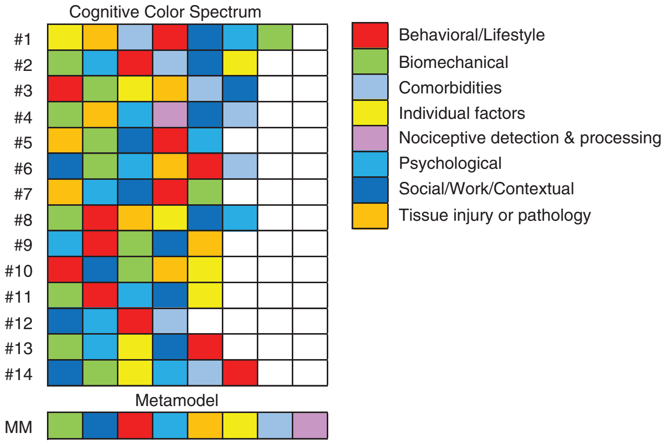

Figure 2. *Cognitive Color Spectra for the individual participants and a metamodel.* Each *category* is ranked by magnitude of the Normalized Sum of Centrality (NSc) for each participant (#1 to #14) and the metamodel (MM). Note that "Biomechanical" *category* has the highest mean NSc for the MM and is the highest ranked *category* for 5 of 14 participants.

conservative[37] and surgical management.[38] This emphasis is exemplified by strong statements such as "PGP is related to non-optimal stability of the pelvic girdle joints" in clinical guidelines.[10] Third, mostly biomechanical factors have been considered to predispose an individual to PGP (eg, falls, repetitive stress, scoliosis, and leg length discrepancy).[2] Fourth, differential diagnosis of PGP has generally involved response to mechanical tests for the SIJ[10,11] and SIJ diagnostic anesthetic blocks.[39,40] Fifth, the greater emphasis on psychosocial rather than biomechanical features in LBP is likely to be largely explained by the limited success of interventions that target the latter.[41] Although it is possible that a mechanical interpretation of PGP is accurate, this does not reflect recent work that has identified associations between several psychological features and persistence of PGP (eg, self-efficacy; anxiety and depression; pain catastrophizing).[42]

Composition of the Metamodel

Consistent with the features of the FCMs, the composition of the metamodel reflected bias toward the "Biomechanics" *category*. This *category* had the highest sum of centrality and included three *components* within the 10 highest centralities ("Poor anatomical/structural characteristics," "Motor impairment," and "Poor posture and alignment"). The related *category* of "Tissue injury or pathology" also ranked highly and included the *component* of "Inflammation" (*category*: Tissue injury or pathology), which had high centrality related to the association between PGP and arthritic conditions including ankylosing spondylitis.

Although "Psychology" was the fourth ranked *category* in the metamodel, the highest ranked individual *component* was the psychological feature of "Cognition (expectations, beliefs, and perceptions concerning pain)," and the centrality attributed to it was almost double that of

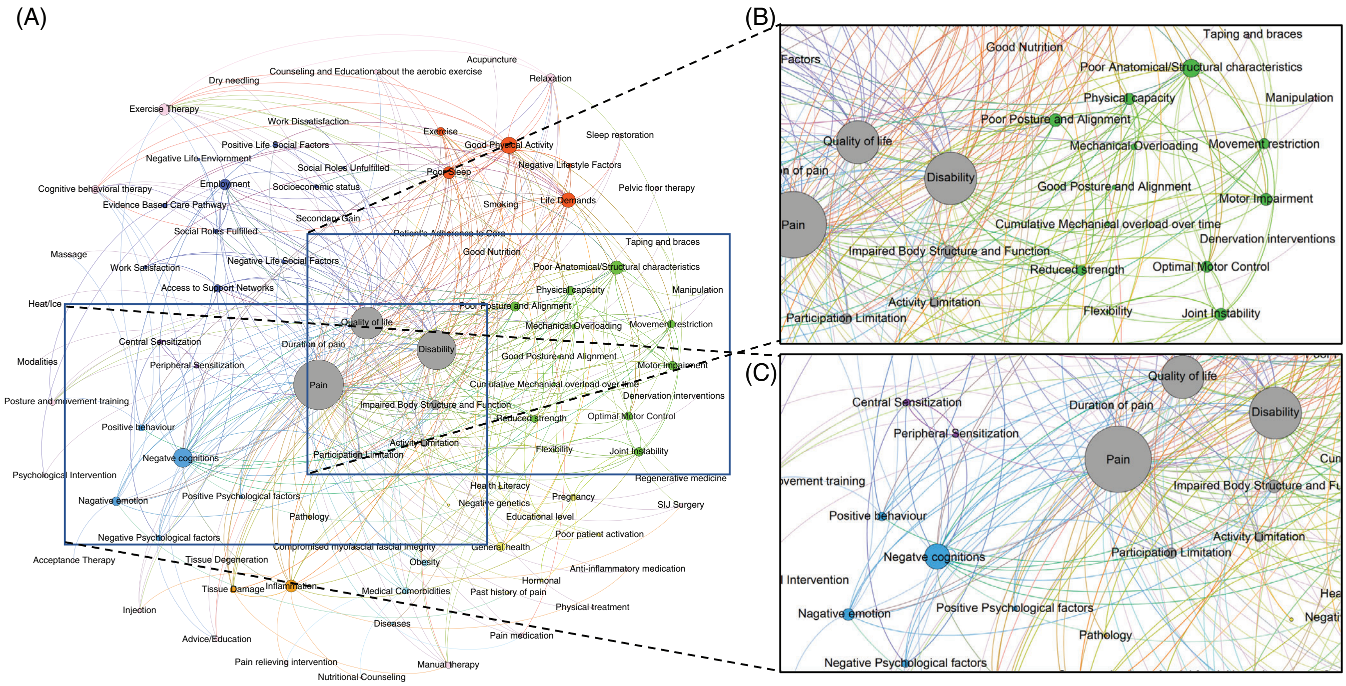

Figure 3. *Metamodel of PGP/SIJ dysfunction.* (A) Complete metamodel of PGP/SIJ dysfunction. All Components and Connections are shown. Categories are identified by color of circles (Components) and outgoing Connections. Size of circles indicates normalized sum of centrality. Treatment components are distributed around the outside of the model. (B) "Biomechanical" components displayed at higher resolutions. (C) "Psychological" components displayed at higher resolution.

Table 4
Centrality of individual *components* in the metamodel

Category	Component	Centrality
Biomechanical	Poor anatomical/structural characteristics	1.012
Biomechanical	Motor impairment	0.982
Biomechanical	Poor posture and alignment	0.876
Biomechanical	Joint instability	0.666
Biomechanical	Strength (reduced)	0.568
Biomechanical	Movement restriction	0.473
Biomechanical	Physical capacity	0.461
Biomechanical	Optimal motor control	0.386
Biomechanical	Cumulative mechanical overload over time	0.250
Biomechanical	Mechanical overloading	0.204
Biomechanical	Good posture and alignment	0.091
Biomechanical	Flexibility	0.057
Social/Work/Contextual factors	Employment	0.870
Social/Work/Contextual factors	Access to support networks	0.838
Social/Work/Contextual factors	Socioeconomic status	0.348
Social/Work/Contextual factors	Evidence-based care pathway	0.257
Social/Work/Contextual factors	Negative life social factors	0.21
Social/Work/Contextual factors	Positive life social factors	0.204
Social/Work/Contextual factors	Work satisfaction	0.200
Social/Work/Contextual factors	Social roles fulfilled	0.179
Social/Work/Contextual factors	Social roles unfulfilled	0.107
Social/Work/Contextual factors	Negative life environment	0.086
Social/Work/Contextual factors	Secondary gain (eg, work environment, motivation, legal)	0.064
Social/Work/Contextual factors	Work dissatisfaction	0.036
Behavioral/Lifestyle	Good physical activity	1.724
Behavioral/Lifestyle	Poor sleep	1.094
Behavioral/Lifestyle	Life demands	0.714
Behavioral/Lifestyle	Exercise	0.547
Behavioral/Lifestyle	Negative lifestyle factors	0.196
Behavioral/Lifestyle	Patient's adherence to care	0.164
Behavioral/Lifestyle	Smoking	0.049
Behavioral/Lifestyle	Good Nutrition	0.021
Psychological	Cognitive (expectations, beliefs & perceptions concerning pain)	3.123
Psychological	Emotional (distress, anxiety and depression)	0.598
Psychological	Negative psychological factors	0.355
Psychological	Behavioural (coping, pain behavior & activity/activity avoidance)	0.346
Psychological	Positive psychological factors	0.136
Tissue injury or pathology	Inflammation	1.079
Tissue injury or pathology	Tissue damage	0.794
Tissue injury or pathology	Compromised myofascial fascial integrity	0.252
Tissue injury or pathology	Pathology	0.100
Tissue injury or pathology	Tissue degeneration	0.043
Individual factors	General health	0.571
Individual factors	Pregnancy	0.380
Individual factors	Poor patient activation (ability to participate in health care)	0.186
Individual factors	Hormonal	0.150
Individual factors	Genetics (negative)	0.096
Individual factors	Health literacy	0.096
Individual factors	Educational level	0.059
Individual factors	Past history of pain	0.043
Comorbidities	Medical comorbidities	0.462
Comorbidities	Overweight (obesity) / BMI	0.344
Comorbidities	Diseases (infections, rheumatoid arthritis, malignancies)	0.121
Nociceptive detection and processing	Central sensitization	0.204
Nociceptive detection and processing	Peripheral sensitization	0.131
Outcomes	Pain	7.923
Outcomes	Disability	5.480
Outcomes	Quality of life	5.258
Outcomes	Activity limitation	1.136
Outcomes	Body structure and function (Impaired)	0.688
Outcomes	Participation limitation	0.582
Outcomes	Duration of pain	0.054

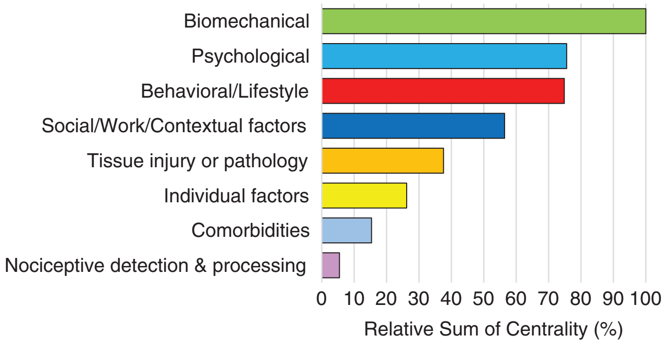

Figure 4. *Relative Sum of Centrality (Sc) for the metamodel.* The Sc values are presented as relative to the "Biomechanical" *category* that had the highest Sc value.

the next ranked *component*. Thus collectively the expert contributors placed weight on psychological issues, but this was focused on a single main feature, in contrast to the multiple separate features in the "Biomechanics" *category*.

Several *components* in the "Behavioral/lifestyle" *category* ranked in the top 10 highest centralities. These were "Poor sleep" and "Good physical activity." In the absence of data in PGP, this is likely to be explained by strong evidence for an association with LBP.[43,44]

Somewhat surprisingly, "Pregnancy" (*category*: Individual) was only ranked 23rd in terms of centrality in the metamodel. This is unexpected considering the high point prevalence of PGP during and after pregnancy (~20%[45]) and the focus on this group in many studies of epidemiology and differential diagnosis.[10] The reason for the limited explicit inclusion of the term "pregnancy" in the metamodel is not clear but is likely explained by inclusion of terms that describe factors associated with pregnancy rather than the term itself.

Interpretation of Relative Efficacy of Treatments Based on the Metamodel

The metamodel generated from the individual contributor FCMs enables investigation of the collective opinion with respect to the expected efficacy of different treatments on "Pain," "Disability," and "Quality of Life." This approach considers the overall weight from all contributors. The treatments predicted to have the greatest expected efficacy differed between outcomes of "Pain," "Disability," and "Quality of Life."

"Injection," "Exercise therapy," and "SIJ surgery" were predicted to have the greatest impact on "Pain." Does this match the best available evidence? "Injection" was used by participants to refer to the intra- or extraarticular injection of several different agents (eg, steroid injections, analgesic agents, prolotherapy, or combination). Although this intervention was considered to have the greatest effect on pain, the efficacy of any injected agent has very limited evidence. Reports of only a few small randomized controlled trials (RCTs; 10-24 participants) with short term follow-up have been published.[46,47] These studies show temporary relief in a subset of individuals, predominantly those with active inflammation (eg, ankylosing spondylitis).[47] Local anesthetic injection is also used for diagnostic purposes.[39] Considering this limited evidence, the strong endorsement of this treatment in the model is somewhat surprising, although it may be explained by the absence of strong evidence for any treatment for PGP. This finding does align with the increasing use of injection procedures in clinical practice.[48]

Evidence for the efficacy of exercise therapy as a treatment for PGP is modest.[10,19] In terms of PGP, exercise therapy has been examined primarily in management of PGP in association with pregnancy, and patients included in trials have commonly not been differentiated from those with LBP, which may be unrelated to PGP. Results are conflicting; some studies show large[37] and long-term effects,[49] whereas others show no effect.[50] Meta-analysis of trials is generally not possible because of heterogeneity of exercise approach and included patient populations. The consensus is that exercise can reduce pain and improve function, but there is little basis to determine which exercise approach is the best.[10] A common approach tested in the literature has been motor control interventions.[19] This concurs with the high centrality in the metamodel of "Motor impairment" and "Poor posture and alignment," which are targeted by these approaches.

Surgery, which includes at least 17 different approaches (most involving fusion).[51] has historically had low evidence.[10] A small number of randomized controlled trials[52,53] and cohort studies are available.[10] Results are variable, with good[52] to poor outcomes reported.[54] Recent trials of a minimally invasive surgical approach have shown promising outcomes for carefully selected individuals with positive response to intra-articular diagnostic anesthetic injections and other diagnostic tests.[52,53] Most authors suggest that surgery is a potential option when nonoperative management has failed.[55] Although failure of nonoperative care has not yet been shown to predict outcome, a recent meta-analysis found better outcomes for the aforementioned carefully selected individuals if they had long duration pain and older age.[56] Outcomes were worse for individuals with a history of opioid use and smoking.

One notable omission from the high-ranking treatments considered to be effective for pain was denervation interventions. Although this might be surprising considering the large literature investigating this approach,[2] it concurs with recent discussion of the limited evidence of clinical efficacy.[57]

Not surprisingly, interventions considered to be effective for "Disability" and "Quality of Life" differed from

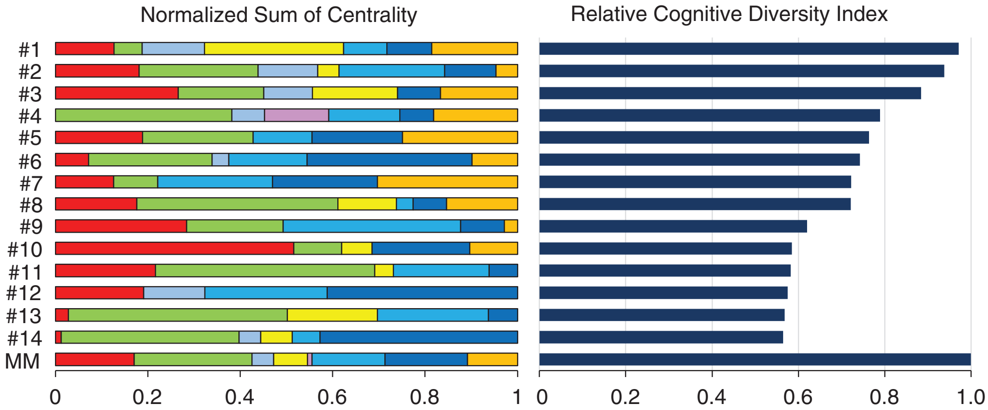

Figure 5. *Normalized Sum of Centrality (NSc) and Cognitive Diversity Index (CDI) for individual participants Fuzzy Cognitive Maps (FCM) and the metamodel (MM).* The order of FCMs is identical to that in Figure 3. Colors in the NSc refer to the *categories* (see Figure 3 for definitions of color). A high CDI indicates that a participant considers *components* across a broad range of *categories* with relatively similar NSc between *categories*. A low CDI indicates that a participant considers *components* across a few *categories* with a bias of NSc to only some *categories*. This analysis does not imply that one model of considering PGP and pain is better or worse, but characterizes the different ways that participants consider the problem.

those for "Pain." Exercise therapy was considered the most effective treatment for "Disability" and "Quality of Life," and there is some evidence for this from RCTs.[10,19] Cognitive behavioral therapy was the second most favored option for both disability and quality of life. Although this appears logical, it has not been investigated in PGP and the perceived potential efficacy of this approach is probably based on evidence from RCTs in LBP.[58,59] Likewise, perceived efficacy of advice/education for PGP is likely to be based on work in LBP,[60] as no RCTs have tested this intervention in patients with PGP. Of interest, "SIJ surgery" was considered the third most likely intervention to impact "Quality of Life." This has some evidence but will likely be relevant for a small subset of patients.[52]

Limitations

By its nature, collaborative modeling aims to summarize the diverse opinions of contributors into a single

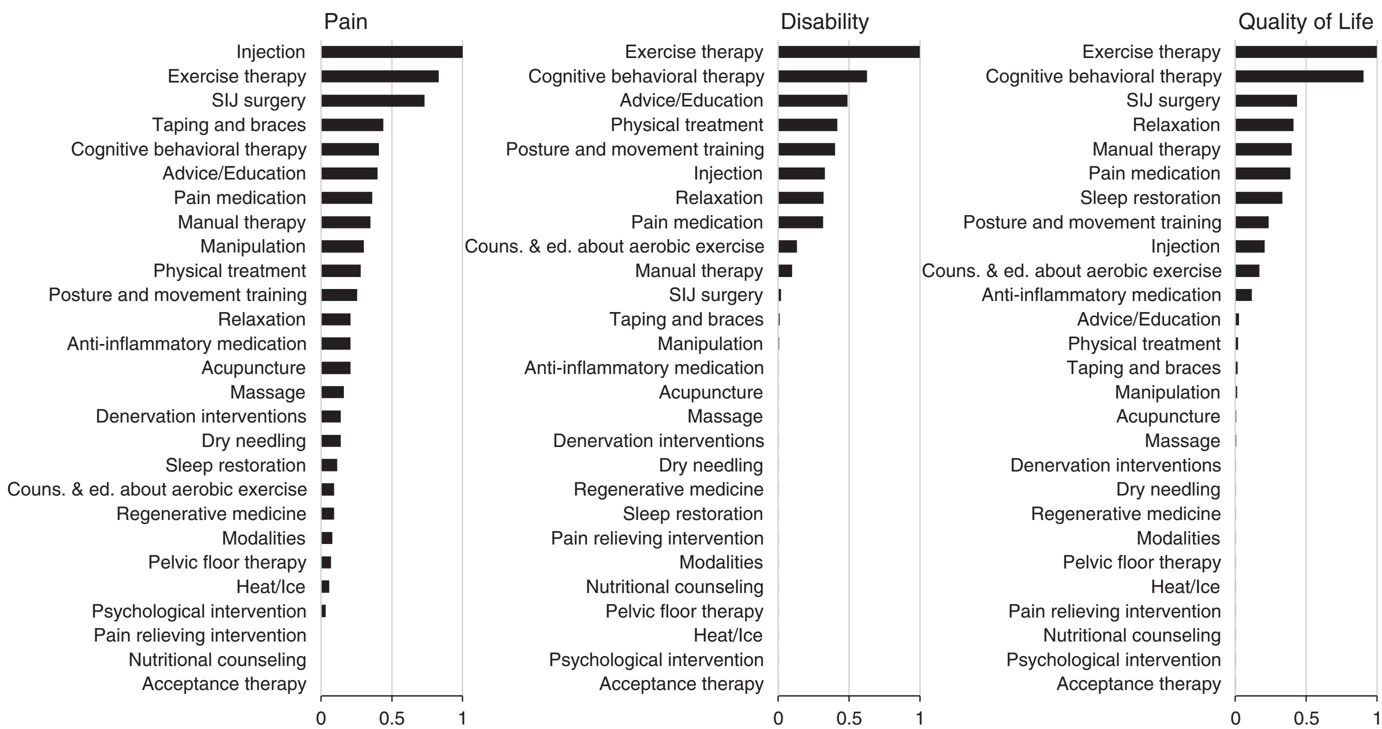

Figure 6. *Metamodel simulations of the effects of various interventions on Pain, Disability, and Quality of Life.* The effects are presented as relative to the most effective intervention and are ranked from the most effective at the top to the least effective at the bottom of each panel.

representative model. This will necessarily involve some simplification and, thus, some limitations. First, the metamodel clusters all presentations of PGP together (eg, ankylosing spondylitis, pregnancy-related conditions, and so on), and although specific treatments may be expected to be effective for specific groups, this cannot be reflected in this model. Second, we collapsed some similar terms from individual FCMs into a smaller group of *components* that were established via extensive consultation with experts (including some who participated in this study) during the development of a collaborative model of LBP.[22] In some cases, the final terminology and grouping may require further consideration. For instance, "Exercise therapy" was nominated as a stand-alone treatment, but several other treatments could also be considered to be forms of exercise (eg, "Posture and movement training," "Counseling and education about aerobic exercise"). Alternatively, it could be argued that exercise therapy is heterogeneous and should be further subdivided into subtypes to better reflect their independent roles. The same issue could be considered for "Injection" and "SIJ surgery," which have multiple forms.

Additional limitations relate to the group of experts who contributed to the model. The group was relatively small and involved mainly individuals from medical and physical therapy backgrounds. Despite the relatively small sample, it has been reported that the number of new variables accumulated per FCM beyond 12 FMCs is relatively small.[26] Although this reflects the bias to these fields in the published literature, greater involvement of individuals from psychology and other disciplines might have changed the centrality of the metamodel *components*. Finally, it is necessary to recognize that this model reflects the opinions of the expert group that we selected and further work is needed to determine whether it reflects opinions more broadly.

Conclusion

This paper presented a collaborative model of PGP. Inspection of the model has provided insight into the complexity of this condition and the relative importance placed by the experts/contributors on different domains in this condition and how this differed from that observed in LBP. The model also exposed a disconnect between perceived relative efficacy of different interventions and the available evidence. The metamodel provides some directions for future research, such as testing some of the proposed *connections* between the *components* and helps in identification of the interventions that should be evaluated as a matter of priority.

Supporting Information

Additional supporting information may be found online in the Supporting Information section at the end of the article.

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