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Diagnostic uncertainty in pediatric chronic pain: Nature, prevalence, and consequences

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Abstract

Introduction: Diagnostic uncertainty (DU), which is the perception that a label or explanation for a patient's health problem is missing or inaccurate, has been linked to distress, anxiety, and difficulty coping among adults with pain. This study examined the prevalence of DU among youth with chronic pain and their parents and the relation of parent and youth DU with youth pain, pain-related constructs, and health-related quality of life.

Methods: Participants included 174 youth with chronic pain ($M_{age}=14.28$ years; 73% female) and one of their parents (91% mothers) recruited from a tertiary-level pediatric chronic pain program in Canada. Youth and parent DU was assessed using a brief measure of three empirically-derived yes/no questions regarding whether the youth and parent had received a clear diagnosis/explanation for their/their child's pain and whether they believed there was something else happening with their/their child's pain which doctors had not yet found. Youth reported on their pain intensity, pain interference, pain catastrophizing, fear of pain, and health-related quality of life.

Results: 31% of youth and 28% of parents experienced DU. 70% of parents and youth were in agreement regarding their experience of DU. Youth DU was linked to higher youth catastrophic thinking about their pain. Parent DU was linked to greater youth pain interference and intensity and lower youth health-related quality of life.

Discussion: DU is experienced by nearly a third of youth with chronic pain and their parents and is linked to worse youth pain, pain catastrophizing, and health-related quality of life.

Summary

Diagnostic uncertainty is common among youth with chronic pain and their parents and is linked to worse youth pain, pain catastrophizing, and quality of life.

Diagnostic uncertainty in pediatric chronic pain: Nature, prevalence, and consequences

Pediatric chronic pain affects 11-38% of youth [12], with approximately 3-5% reporting significant disability [7;8]. In the absence of underlying organic pathology or positive test results, many youth receive a primary diagnosis of ‘chronic pain’ [4] or ‘Primary Pain Disorder’ [24]. As a consequence, youth and their parents may experience uncertainty regarding the diagnosis, prognosis, and treatment [15]. Diagnostic uncertainty (DU), the perception that a label or explanation for a patient’s health problem is missing or inaccurate, is associated with higher levels of anxiety, depression, pain intensity [27], and disability [5] among adults with chronic primary pain. Research suggests that over 40% of adults with chronic low back pain experience DU [26]. However, the prevalence and impact of DU in the context of pediatric chronic primary pain is relatively unknown.

Nearly 40% of parents of youth with chronic primary pain report not accepting or ‘buying in’ to their child’s diagnosis [18], suggesting that the prevalence of DU among parents of youth with chronic pain may be consistent with adult literature. In a qualitative examination of DU among 20 youth with chronic primary pain and their parents, DU was characterized by a relentless search for the ‘right’ diagnosis, which may limit acceptance of a diagnosis and delay or prevent engagement in treatment [15]. A recent topical review proposed that research should examine the prevalence of DU among youth with chronic pain and their parents, and its influence on treatment and pain outcomes [23]. A brief measure of DU was developed for adults with chronic pain [23;25;26], but has never been used among youth with chronic pain and their parents.

This study aimed to 1) describe the prevalence and nature of DU among youth with chronic pain and their parents, 2) compare youth pain (intensity and interference), pain-related

constructs (pain catastrophizing, fear of pain) and health-related quality of life (HRQoL) in youth with chronic pain who do versus do not report DU and in parents of youth who do versus do not report DU. We hypothesized that prevalence rates of DU would parallel adult literature and that youth and parent DU would be related to worse youth pain, pain-related constructs, and HRQoL.

Methods

The current cross-sectional survey is part of the Pain and Mental Health in Youth study (PATH). The current aims are distinct from previously published papers on this study [14;15;16;22]. The study was approved by the institutional health Research Ethics Board. Participants included 174 youth with chronic pain ($M_{age}=14.28$ years; 73% female, range=10-18 years) and one of their parents (91% mothers) recruited from a tertiary-level pediatric chronic pain program in Canada. Youth were enrolled from headache (67%), complex pain (28%), or abdominal pain (5%) clinics. Youth were eligible for the study if they were between 10-18 years of age and identified as having chronic pain (i.e., pain ≥ 3 months [13]) without an underlying disease (e.g., cancer). Youth with a diagnosis of a severe cognitive disability or developmental disorder were excluded. Parents and youth consented and completed self-report questionnaires using Research Electronic Data Capture (REDCap) [6].

Measures

Parent and youth diagnostic uncertainty. DU was assessed using a brief measure consisting of three empirically derived categorial questions, which were constructed from a study investigating DU among adults with low back pain [25;26]. This is the first study to use this measure since it was adjusted for use with any chronic pain condition [23]. Youth and parents responded yes/no to 3 questions presented in Table 1. Research has demonstrated that individuals with chronic pain often simultaneously report having received a clear diagnosis, while believing

that something else is happening with their pain [27]. Thus, question 3 is particularly informative in terms of DU and has been linked to adverse outcomes among adults with chronic pain [23;26;27]. DU was therefore operationalized as youth and parents who responded ‘yes’ to question 3. If youth or parents responded yes to questions 1 or 2, they were asked whether or not they agreed with the diagnosis or explanation provided to them.

Youth pain. Youth reported on their pain characteristics using the valid and reliable Pain Questionnaire [21]. Youth reported their average pain intensity in the past seven days using a validated 11-point numeric rating scale (0=“no pain”, 10=“worst pain possible”) [1;30]. Youth completed the Pain Interference Subscale of the Patient-Reported Outcomes Measurement Information System (PROMIS) Pediatric Profile-25 [9;11;17;19;20].

Youth pain-related constructs. The Pain Catastrophizing Scale-Child version (PCS-C) [3] was used to assess catastrophic thoughts about child pain. The Fear of Pain Questionnaire, child report (FOPQ-C), was used to assess pain-related fears and avoidance behaviours [28].

Youth health-related quality of life. HRQoL was assessed using the Pediatric Quality of Life Short Form (PedsQL-SF) [2].

Results

Tables 2 and 3 present sample characteristics and key variable associations. Girls reported significantly higher pain interference than boys ($p<.05$). Older youth reported higher pain interference ($r=0.17$, $p<.05$), fear of pain ($r=0.24$, $p<.01$), and lower HRQoL ($r=-0.29$, $p<.01$) than younger youth.

Participants’ responses to the DU questions are summarized in Table 4. Nearly a third of youth (31%) and parents (28%) believed something else was happening with their/their child’s pain which the doctors had not yet found. 21% of youth and 17% of parents who reported that

they had received a clear diagnosis also reported thinking something else was happening with their/their child's pain. 70% of parents and youth concurred regarding their DU (i.e., both responded yes or no to question 3); 16% of parents and 14% of youth reported being uncertain while their child or parent was certain, respectively.

Table 5 presents descriptive statistics and results of ANCOVA analyses. Controlling for youth age and gender, youth who experienced DU reported significantly higher pain catastrophizing than youth who did not experience DU. Youth whose parents experienced DU reported significantly greater pain interference, greater pain intensity, and lower HRQoL than youth whose parents did not experience DU.

Discussion

This is the first quantitative investigation of DU among a large sample of youth with chronic pain and their parents. Nearly a third of youth with chronic pain and their parents experienced DU. Consistent with findings among adults with low back pain [26], a substantial minority of youth and parents experienced DU despite reporting that they received a clear diagnosis for their pain. Serbic and Pincus postulated that such findings may indicate beliefs that a diagnosis may be correct but does not fully capture the cause of the pain [26]. In the current study, most parents and youth who reported receiving a diagnosis also reported *agreeing* with that diagnosis. Yet, many families, who are imbedded in a tertiary level chronic pain program for primary pain disorders, continue to believe something undiagnosed is causing the pain. This finding alludes to the perception that the diagnosis provided did not fully explain the youth's pain and what was lacking was the core etiology. This has important implications for clinical encounters when a chronic pain diagnosis is provided, suggesting that the diagnosis itself is not sufficient to allay fears and beliefs that something unknown is causing the pain. Qualitative

research with youth with chronic pain and their parents has revealed that many parents and youth express needing further explanation for their pain [10;15]. Indeed, a chronic pain diagnosis is often perceived as a diagnosis of exclusion and to not reflect a true understanding of the cause of the pain [15]. Further research is needed to understand how clinicians could tailor their explanation of a chronic pain diagnosis in a way that would reduce DU.

This study is the first to investigate the relation of youth and parent DU with youths' pain, pain-related constructs, and HRQoL. Whereas youth DU was linked to higher youth catastrophic thinking about their pain, it was *parent* DU that was linked to greater youth pain interference and intensity and lower HRQoL. In line with the interpersonal fear avoidance model of pain, uncertainty may fuel parental distress and protective responses around child pain, which may facilitate youth avoidance behaviours and influence youth pain interference [16;29]. Further longitudinal research is needed to understand how DU unfolds over time, its temporal relations with outcomes, and what factors during diagnosis and treatment influence DU. Moreover, the direction of the association between youth pain and parent DU remains unknown—that is, whether having a child with worse pain exacerbates parent DU or vice versa.

A limitation of the current study is the dichotomous nature of the DU questions, which may not capture the varying intensity and nuances of DU [15]. DU may fluctuate over time depending on where youth are in their pain and treatment journey, which was not assessed in the current study. Further, the current data is based on self-report; thus, the information provided to families regarding diagnosis is unknown. Future research should examine how DU differs based on chronic pain diagnosis and explanation (e.g., etiological versus primary pain). Nevertheless, our findings demonstrate that DU is a common experience among youth with chronic pain and their parents and is linked to worse pain, pain-related constructs, and HRQoL.

References

- [1] Castarlenas E, Jensen MP, von Baeyer CL, Miro J. Psychometric properties of the Numerical Rating Scale to assess self-reported pain intensity in children and adolescents: A systematic review. *Clin J Pain* 2017;33(4):376-383.
- [2] Chan KS, Mangione-Smith R, Burwinkle TM, Rosen M, Varni JW. The PedsQL: Reliability and validity of the short-form generic core scales and Asthma Module. *Med Care* 2005;43(3):256-265.
- [3] Crombez G, Bijttebier P, Eccleston C, Mascagni T, Mertens G, Goubert L, Verstraeten K. The child version of the pain catastrophizing scale (PCS-C): A preliminary validation. *Pain* 2003;104(3):639-646.
- [4] Eccleston C, Malleson P. Managing chronic pain in children and adolescents. We need to address the embarrassing lack of data for this common problem. *BMJ* 2003;326(7404):1408-1409.
- [5] Geisser ME, Roth RS. Knowledge of and agreement with chronic pain diagnosis: Relation to affective distress, pain beliefs and coping, pain intensity, and disability. *J Occup Rehabil* 1998;8(1):73-88.
- [6] Harris PA, Taylor R, Thielke R, Payne J, Gonzalez N, Conde JG. Research electronic data capture (REDCap)--a metadata-driven methodology and workflow process for providing translational research informatics support. *J Biomed Inform* 2009;42(2):377-381.
- [7] Hechler T, Dobe M, Zernikow B. Commentary: A worldwide call for multimodal inpatient treatment for children and adolescents suffering from chronic pain and pain-related disability. *J Pediatr Psychol* 2010;35(2):138-140.

- [8] Huguet A, Miro J. The severity of chronic pediatric pain: An epidemiological study. *J Pain* 2008;9(3):226-236.
- [9] Irwin DE, Stucky BD, Thissen D, Dewitt EM, Lai JS, Yeatts K, Varni JW, DeWalt DA. Sampling plan and patient characteristics of the PROMIS pediatrics large-scale survey. *Qual Life Res* 2010;19(4):585-594.
- [10] Jordan AL, Eccleston C, Osborn M. Being a parent of the adolescent with complex chronic pain: An interpretative phenomenological analysis. *Eur J Pain* 2007;11(1):49-56.
- [11] Kashikar-Zuck S, Carle A, Barnett K, Goldschneider KR, Sherry DD, Mara CA, Cunningham N, Farrell J, Tress J, DeWitt EM. Longitudinal evaluation of patient-reported outcomes measurement information systems measures in pediatric chronic pain. *Pain* 2016;157(2):339-347.
- [12] King S, Chambers CT, Huguet A, MacNevin RC, McGrath PJ, Parker L, MacDonald AJ. The epidemiology of chronic pain in children and adolescents revisited: A systematic review. *Pain* 2011;152(12):2729-2738.
- [13] Merskey H, Bogduk N. Classification of chronic pain: Descriptions of chronic pain syndromes and definitions of pain terms. Task Force on Taxonomy of the International Association for the Study of Pain 1994.
- [14] Neville A, Griep Y, Palermo TM, Vervoort T, Schulte F, Yeates KO, Sumpton JE, Mychasiuk R, Noel M. A 'dyadic dance': Pain catastrophizing moderates the daily relationships between parent mood and protective responses and child chronic pain. *Pain* 2020.
- [15] Neville A, Jordan A, Beveridge JK, Pincus T, Noel M. Diagnostic uncertainty in youth with chronic pain and their parents. *J Pain* 2019.

- [16] Neville A, Kopala-Sibley DC, Soltani S, Asmundson GJ, Jordan A, Carleton RN, Yeates KO, Schulte F, Noel M. A longitudinal examination of the interpersonal fear avoidance model of pain: the role of intolerance of uncertainty. *Pain* 2020.
- [17] Neville A, Soltani S, Pavlova M, Noel M. Unravelling the relationship between parent and child PTSD and pediatric chronic pain: The mediating role of pain catastrophizing. *J Pain* 2018;19(2):196-206.
- [18] Noel M, Beals-Erickson SE, Law EF, Alberts NM, Palermo TM. Characterizing the pain narratives of parents of youth with chronic pain. *Clin J Pain* 2016;32(10):849-858.
- [19] Noel M, Vinall J, Tomfohr-Madsen L, Holley AL, Wilson AC, Palermo TM. Sleep mediates the association between PTSD symptoms and chronic pain in youth. *J Pain* 2018;19(1):67-75.
- [20] Noel M, Wilson AC, Holley AL, Durkin L, Patton M, Palermo TM. Posttraumatic stress disorder symptoms in youth with vs without chronic pain. *Pain* 2016;157(10):2277-2284.
- [21] Palermo TM, Witherspoon D, Valenzuela D, Drotar DD. Development and validation of the Child Activity Limitations Interview: A measure of pain-related functional impairment in school-age children and adolescents. *Pain* 2004;109(3):461-470.
- [22] Pavlova M, Kopala-Sibley DC, Nania C, Mychasiuk R, Christensen J, McPeak A, Tomfohr-Madsen L, Katz J, Palermo TM, Noel M. Sleep disturbance underlies the co-occurrence of trauma and pediatric chronic pain: A longitudinal examination. *Pain* 2020.
- [23] Pincus T, Noel M, Jordan A, Serbic D. Perceived diagnostic uncertainty in pediatric chronic pain. *Pain* 2018.
- [24] Schechter NL. Functional pain: Time for a new name. *JAMA Pediatr* 2014;168(8):693-694.

- [25] Serbic D, Pincus T. Chasing the ghosts: The impact of diagnostic labelling on self-management and pain-related guilt in chronic low back pain patients. *J Pain Manag* 2013;1(6):25-35.
- [26] Serbic D, Pincus T. Diagnostic uncertainty and recall bias in chronic low back pain. *Pain* 2014;155(8):1540-1546.
- [27] Serbic D, Pincus T, Fife-Schaw C, Dawson H. Diagnostic uncertainty, guilt, mood, and disability in back pain. *Health Psychol* 2016;35(1):50-59.
- [28] Simons LE, Sieberg CB, Carpino E, Logan D, Berde C. The Fear of Pain Questionnaire (FOPQ): Assessment of pain-related fear among children and adolescents with chronic pain. *J Pain* 2011;12(6):677-686.
- [29] Simons LE, Smith A, Kaczynski K, Basch M. Living in fear of your child's pain: The Parent Fear of Pain Questionnaire. *Pain* 2015;156(4):694-702.
- [30] von Baeyer CL, Spagrud LJ, McCormick JC, Choo E, Neville K, Connelly MA. Three new datasets supporting use of the Numerical Rating Scale (NRS-11) for children's self-reports of pain intensity. *Pain* 2009;143(3):223-227.

Table 1
Brief measure of perceived diagnostic uncertainty

Questions
<hr/>
Youth
1. I have been given a clear label/diagnosis for my pain
2. I have been given a clear explanation about why I have pain
3. I think there is something else happening with my pain, which the doctors have not found out about yet.
Parent
1. My child has been given a clear label/diagnosis for their pain
2. My child has been given a clear explanation about why they have pain
3. I think there is something else happening with my child's pain, which the doctors have not found out about yet.

Table 2
Socio-demographic and pain characteristics of the sample

Sample Characteristics	<i>N</i> = 174
Child age (<i>M</i> years, <i>SD</i>)	14.28(2.22)
Child gender (% female)	73.0
Parent gender (% female)	91.3
Child's Ethnicity (%)	
White	79.3
Two or more ethnicities	8.0
Other	4.2
Black	1.7
Arab/West Asian	1.7
South Asian	1.7
Aboriginal	1.1
Latin American	1.1
Filipino	0.6
Did not want to answer	0.6
Annual household income (%)	
<\$10,000-\$29,999	5.7
\$30,000-\$59,999	10.3
\$60,000-\$89,999	12.1
>\$90,000	57.5
Did not want to answer	9.2
No income reported	5.2
Pain locations (%)	
Multiple locations reported	43.7
Head	72.4
Other	24.7
Muscles and joints	23.0
Stomach	20.1
Legs	12.6
Chest	10.9
Child pain intensity (<i>M</i> out of 10, <i>SD</i>)	5.53(1.84)
Child pain duration (<i>M</i> years, <i>SD</i>)	3.20(3.11)

Table 3
Correlations among variables of interest

Variable	1	2	3	4	5
(1) Youth pain intensity	1	0.50 [‡]	0.39 [‡]	0.33 [‡]	-0.39 [‡]
(2) Youth Pain interference		1	0.48 [‡]	0.50 [‡]	-0.60*
(3) Youth pain catastrophizing			1	0.77 [‡]	-0.60 [‡]
(4) Youth fear of pain				1	-0.65 [‡]
(5) Youth quality of life					1

Note. ‡ $p < .01$; * $p < .05$

Table 4
Participant responses to diagnostic uncertainty questions

	Yes n(%)		No n(%)		Total n	
	Youth	Parents	Youth	Parents	Youth	Parents
I/My child have/has been given a clear label/diagnosis for my/their pain	72(42.6)	79(46.2)	97(57.4)	92(53.8)	169	171
I agree with this label/diagnosis	70(97.2)	76(96.2)	2(2.8)	3(3.8)	72	79
I/My child have/has been given a clear explanation about why I/they have pain	42(24.9)	73(42.9)	127(75.1)	97(57.1)	169	170
I agree with this explanation	39(92.9)	68(94.4)	3(7.1)	4(5.6)	42	72
I think there is something else happening with my/my child's pain, which the doctors have not found out about yet.	51(30.5)	47(27.5)	116(69.5)	124(72.5)	167	171

Table 5

ANCOVA results controlling for youth age and gender

Variable	<i>N</i>	Uncertain <i>M (SD)</i>	Certain <i>M (SD)</i>	<i>df</i>	<i>F</i>	<i>P</i>
Parent DU						
Youth pain interference	165	57.83 (9.07)	54.06 (8.90)	1,161	4.23	.04
Youth pain intensity	165	6.09 (1.55)	5.27 (1.86)	1,161	5.81	.02
Youth Catastrophizing	164	21.67 (12.44)	18.01 (12.08)	1,160	2.38	.13
Youth fear of pain	160	35.91 (22.65)	28.50 (19.13)	1,156	3.48	.06
Youth health-related quality of life	164	57.86 (19.04)	68.14 (19.10)	1,160	7.44	.01
Youth DU						
Youth pain interference	163	56.04 (9.11)	54.49 (9.16)	1,159	.88	.35
Youth pain intensity	163	5.90 (1.68)	5.30 (1.86)	1,159	3.81	.05
Youth Catastrophizing	162	22.60 (12.50)	17.42 (11.70)	1,158	6.06	.02
Youth fear of pain	159	34.90 (18.87)	28.40 (20.55)	1,155	3.45	.07
Youth health-related quality of life	162	61.25 (19.56)	67.32 (19.01)	1,158	3.37	.07