

Mother-reported pain experience between ages 7 and 10: A prospective study in a population-based birth cohort

Raquel Lucas^{1,2}   | Makram Talih²  | Teresa Monjardino² | Susana Guimarães² | Henrique Barros^{1,2} 

¹Departamento de Ciências da Saúde Pública e Forenses e Educação Médica, Faculdade de Medicina, Universidade do Porto, Porto, Portugal

²EPIUnit (Epidemiology Research Unit), Instituto de Saúde Pública, Universidade do Porto, Porto, Portugal

Correspondence

Raquel Lucas, Instituto de Saúde Pública, Universidade do Porto, Porto, Portugal.
Email: rluca@med.up.pt

Funding information

This study was funded by the European Regional Development Fund (ERDF), through COMPETE 2020 Operational Programme 'Competitiveness and Internationalisation' together with national funding from the Foundation for Science and Technology (FCT)—Portuguese Ministry of Science, Technology and Higher Education—through the projects "STEPACHE—The paediatric roots of amplified pain: from contextual influences to risk stratification" (POCI-01-0145-FEDER-029087, PTDC/SAU-EPI/29087/2017), and "HIneC: When do health inequalities start? Understanding the impact of childhood social adversity on health trajectories from birth to early adolescence" (POCI-01-0145-FEDER-029567, PTDC/SAU-PUB/29567/2017). This work was also supported by the Epidemiology Research Unit—Instituto de Saúde Pública, Universidade do Porto (EPIUnit) (POCI-01-0145-FEDER-006862; UID/DTP/04750/2019), by Administração Regional de Saúde Norte (Regional Department of the Portuguese Ministry of Health) and Calouste Gulbenkian Foundation.

Abstract

Background: Trajectory studies suggest considerable stability of persistent or recurrent pain in adolescence. This points to the first decade of life as an important aetiologic window for shaping future pain, where the potential for prevention may be optimised.

Objectives: We aimed to quantify changes in mother-reported pain experience in children between ages 7 and 10 and describe clusters of different pain experiences defined by complementary pain features.

Methods: We conducted a prospective study using data from 4036 Generation XXI birth cohort participants recruited in 2005–06. Pain history was reported by mothers at ages 7 and 10 using the Luebeck pain screening questionnaire. We tracked changes in six pain features over time using relative risks (RRs) and their 95% confidence intervals (95% CIs). Clusters were obtained using the k-medoids algorithm.

Results: The risk of severe pain at age 10 increased with increasing severity at age 7, with RRs ranging from 2.18 (95% CI 1.90, 2.50) for multisite to 4.43 (95% CI 3.19, 6.15) for high frequency pain at age 7. A majority of children (59.4%) had transient or no pain but two clusters included children with stable recurrent pain ($n = 404$, 10.2% of the sample). One of those ($n = 177$) was characterised by higher probabilities of multisite pain (74.6% and 66.7% at ages 7 and 10, respectively), with psychosocial triggers/contexts (59.3% and 61.0%) and daily-living restrictions (72.2% and 84.6%). Most children in that cluster (58.3%) also self-reported recent pain at age 10 and had more frequent family history of chronic pain (60.5%).

Conclusions: All pain features assessed tracked with a positive gradient between ages 7 and 10, arguing for the significance of the first decade of life in the escalation of the pain experience. Multisite pain and psychosocial attributions appeared to be early markers of more adverse pain experiences.

KEYWORDS

birth cohort, course of pain, maternal report, paediatric pain, population-based, prospective study

Social media quote: In a birth cohort, the experience of pain escalated between child ages 7 and 10 as reported by mothers. Multisite pain and psychosocial attributions appeared to be distinctive markers of more adverse pain experiences.

This is an open access article under the terms of the Creative Commons Attribution-NonCommercial-NoDerivs License, which permits use and distribution in any medium, provided the original work is properly cited, the use is non-commercial and no modifications or adaptations are made.

© 2020 The Authors. *Paediatric and Perinatal Epidemiology* published by John Wiley & Sons Ltd.



1 | BACKGROUND

Persistent or recurrent pain in children is frequently reported in the absence of an identifiable organic cause.¹ Its clinical significance can be tied to the establishment of lifelong pain phenotypes.² Repeated pain measurements in otherwise healthy youth may separate manifestations of a heightened pain propensity in response to evolving triggers or sequential sensitising steps in the same causal path.^{3,4} From both perspectives—causal or predictive—early identification of children with increased susceptibility to chronic pain trajectories is a cornerstone of current pain research.⁵

Although most evidence on paediatric pain prognosis has been obtained in clinical settings,⁶ a few prospective studies have described the course of pain in cohorts selected independently of initial symptoms.⁷⁻⁹ In the United States, four to six different trajectories were identified including a majority of adolescents without relevant complaints, a considerable proportion with transient pain, and a smaller group with persistent pain.⁷ Four low back pain clusters (low, increasing, decreasing, and high level) were also described in Australia,⁸ and a similar typology was found for headache in German girls.⁹ Overall, trajectory studies argue for a considerable stability of persistent or recurrent pain as early as in adolescence. This points to the first decade of life as an important aetiologic window for shaping future pain. Knowledge on how different features of pain evolve in this stage can help identify early markers for chronic pain phenotypes. In addition to classic symptom attributes, causal attributions may be particularly important, since they seem to shape health-related distress and prognosis.¹⁰⁻¹²

Prospective birth cohorts are privileged settings to trace chronic pain onset. Although pain is by definition an individual experience, parents are a key source of information in the first decade of life. As in clinical paediatrics, where it remains essential for anamnesis, parent-reported pain can be confronted with the child's own account, both as proxy for and determinant of the child's experience. Using data from evaluations at 7 and 10 years of age of a population-based birth cohort, we aimed to characterise the course of pain as reported by mothers. Specifically, we aimed to quantify changes in pain experience between ages 7 and 10 and cluster children according to features experienced: symptom attributes (number of sites, duration, frequency, and intensity), causal attributions, and resulting restrictions.

2 | METHODS

2.1 | Cohort selection

The present study is based on the Generation XXI birth cohort, described in detail elsewhere.^{13,14} Briefly, mothers who gave birth to liveborn children with gestational age over 23 weeks in 2005-06 were recruited up to 72 hours after delivery from the five public maternities that covered the metropolitan area of Porto, Portugal. A total of 91.4% agreed to participate, yielding an initial cohort of 8647 children. Interval follow-ups were conducted at 4, 7, and 10 years of age.

Synopsis

Study question

We wanted to quantify changes in mother-reported pain experience in children between ages 7 and 10 and to describe clusters of different pain experiences defined by complementary pain features.

What's already known

The aetiology of chronic pain throughout life is thought to trace back to paediatric ages. Trajectory studies in the second decade of life suggest considerable stability of persistent or recurrent pain already in adolescence.

What this study adds

We found that pain experiences tracked with a positive gradient between ages 7 and 10, suggesting that this may be an important period in the establishment and escalation of the pain experience. Multisite pain and psychosocial attributions appeared to be distinctive early markers of adverse pain experiences.

Each follow-up included face-to-face interviews to collect information about the family (demographic and socio-economic characteristics, family structure, and parental medical history) and index child (medical history, physical symptoms, and health-related behaviours). A comprehensive physical examination of the child was also conducted.

2.2 | Ethics approval

The protocol complied with the Declaration of Helsinki and was approved by the joint Ethics Committee of the São João Hospital and University Centre and the University of Porto Medical School. Families were considered participant if legal guardians signed a written informed consent form and children provided oral assent.

2.3 | Maternal report of child's pain

A Portuguese version of the Luebeck pain screening questionnaire was used.¹⁵ The first (screening) question was "Did your child complain of pain in the last 3 months?" If the answer was affirmative, the mother was asked to select the anatomical sites where the child felt pain and indicate the site of the principal pain according to her assessment. The number of sites was categorised as 0, 1-2, or ≥ 3 sites. If the principal pain had occurred only once in the three months prior to interview, no further questions were asked. Mother-reported

occurrence of pain was categorised as none, one episode, or recurrent (two or more episodes).

If the principal pain was recurrent, the mother was asked about: duration, recoded as short (≤ 3 months), medium (4–12 months), or long (> 12 months); frequency, recoded as low (at most once a month), medium (more than once a month, but at most once a week), or high (more than once a week); and intensity, reported using both the original six-point Faces Pain Scale (FPS)¹⁶ and a visual analog scale (VAS)¹⁷ ranging from 0 (“hardly noticeable pain”) to 100 mm (“strongest conceivable pain”). Mother-reported intensity was recoded as low (FPS 1–3 and VAS < 30), medium (FPS 1–3 and $30 \leq \text{VAS} < 60$, or FPS 4 and VAS < 60), or high (FPS 5–6 or VAS ≥ 60). We combined FPS and VAS to increase sensitivity for more adverse experiences, particularly in respondents with missing information in one of the scales (eg, in case of lower literacy).

The mother was asked about whether the child needed to visit a doctor because of the recurrent principal pain and about impact on the child’s daily living (missing school, not being able to meet with friends or engage in leisure time activities, having trouble eating or sleeping). Pain triggers and context of first episode were also reported. We grouped pain attributions as psychological, somatic, or normalising/situational.¹² Since attributional styles that tend towards psychological causes are associated with the highest distress,¹⁰ pain reports were dichotomised as any psychosocial (any mention of interpersonal or psychological distress; see footnote in Table 1) versus no psychosocial trigger/context reported. Finally, if any of the child’s parents or siblings were reported to suffer from chronic pain, then we considered that the child had a family history of chronic pain.

We are not aware of any previous validation of the Luebeck pain screening questionnaire in Portugal. The instrument collects standard elements of anamnesis originally analysed at face value,¹⁵ and therefore, we used each pain attribute as a separate input in the analyses.

2.4 | Pain report by child

As a brief measure of criterion validity, children aged 10 were asked “During the last week, did you feel pain in any region of your body?” Those who answered affirmatively were asked to indicate the regions affected in a body chart. Children reported the intensity of their most disturbing pain using a six-point FPS.

2.5 | Statistical analyses

All mother-reported pain features were tabulated by sex at birth and evaluation wave. The magnitude and precision of comparisons between waves were evaluated using prevalence ratios (PRs) and their 95% confidence intervals (95% CIs).

To track adverse pain experience, conditional probabilities of mothers reporting each pain feature category at age 10 given the report of that category at age 7 were tabulated. The magnitude and precision of associations between each feature at age 7 and

reporting the highest severity category at age 10 were estimated using relative risks (RRs) and their 95% CIs.

Children were grouped according to their sex and the mother-reported features of their pain episode(s) at ages 7 and 10. The k-meoids algorithm was used to determine optimal cluster centres and membership for the 3954 children with available data at both ages.¹⁸ The number of clusters, $k = 11$, and initial cluster centres were based on the dendrogram of a preliminary hierarchical clustering computed by complete linkage on the subsample with complete pain features ($n = 3862$).¹⁹ Gower distance was used as the dissimilarity measure between index children.²⁰

The number and composition of clusters obtained agreed with results (not shown) from model-based (mixtures of multinomial distributions) and (nonparametric) density-based clustering, which provided some validation of the final set of clusters and was consistent with our understanding of the interrelation among the pain features.²¹

2.6 | Missing data

Of the 4036 children in our analytic sample, 82 had missing data on the features of principal pain in either wave 7 or 10, as those were only collected for recurrent pain due to questionnaire design. Another 92 children had missing data on at most one feature, which we imputed according to the estimated cell probabilities in the age- and sex-specific contingency table formed by the six pain features. As a result, 3954 children were retained for clustering.

2.7 | Nonparticipation and sensitivity analysis

Due to nonparticipation and data-related exclusions (Figure 1), no information on pain experience at ages 7 or 10 was available for 2428 children of the initial cohort. These children were born to younger mothers, with lower educational level, and higher parity, but had similar birthweight and gestational age (Table S1). Mothers with lower educational level tended to report a more severe pain experience for their children, (Table S2). However, this observation was not robust across features, and after stratifying the associations between pain features at ages 7 and 10 by maternal education, our findings were mainly unchanged (Figure S1). None of the other baseline characteristics tested were clearly associated with pain features.

All statistical analyses were performed in R (version 3.6.1),²² except Figure 3, which was built in Stata (version 15.1).

3 | RESULTS

In our sample of 4036 children (49.1% girls), mother-reported prevalence of pain in the three months prior to interview was 45.4% at age 7 and 58.1% at age 10 (breakdown by sex in Table 1). Three or more pain sites were reported for 20.6% of children at



TABLE 1 Distribution, n (%), of mother-reported occurrence of pain, number of sites, principal pain site, and features of the principal recurrent pain at ages 7 and 10, by sex of the child, in the three months prior to interview

	Age 7			Age 10		
	Boys	Girls	Total	Boys	Girls	Total
Pain occurring in past three months?						
None	1160 (56.5)	1036 (52.2)	2196 (54.4)	891 (43.4)	725 (36.6)	1616 (40.0)
One episode	363 (17.7)	332 (16.7)	695 (17.2)	650 (31.7)	634 (32.0)	1284 (31.8)
Recurrent (two or more episodes)	523 (25.5)	613 (30.9)	1136 (28.1)	484 (23.6)	578 (29.1)	1062 (26.3)
Multisite pain						
0 sites	1161 (56.6)	1036 (52.2)	2197 (54.4)	891 (43.4)	726 (36.6)	1617 (40.1)
1-2 sites	521 (25.4)	486 (24.5)	1007 (25.0)	784 (38.2)	793 (40.0)	1577 (39.1)
≥3 sites	371 (18.1)	461 (23.2)	832 (20.6)	378 (18.4)	464 (23.4)	842 (20.9)
Principal pain site ^a						
Abdomen	199 (9.7)	221 (11.1)	420 (10.4)	177 (8.6)	238 (12.0)	415 (10.3)
Head	185 (9.0)	220 (11.1)	405 (10.0)	253 (12.3)	264 (13.3)	517 (12.8)
Lower limbs	223 (10.9)	172 (8.7)	395 (9.8)	311 (15.1)	214 (10.8)	525 (13.0)
Ears and throat	98 (4.8)	149 (7.5)	247 (6.1)	123 (6.0)	156 (7.9)	279 (6.9)
Teeth and mouth	89 (4.3)	76 (3.8)	165 (4.1)	70 (3.4)	54 (2.7)	124 (3.1)
Pelvis and hips	31 (1.5)	37 (1.9)	68 (1.7)	48 (2.3)	133 (6.7)	181 (4.5)
Back, neck, and shoulders	15 (0.7)	29 (1.5)	44 (1.1)	42 (2.0)	60 (3.0)	102 (2.5)
Thorax, chest, and breasts	22 (1.1)	20 (1.0)	42 (1.0)	48 (2.3)	64 (3.2)	112 (2.8)
Upper limbs	7 (0.3)	4 (0.2)	11 (0.3)	18 (0.9)	12 (0.6)	30 (0.7)
Other site(s) or site unspecified	15 (0.7)	11 (0.6)	26 (0.6)	34 (1.7)	14 (0.7)	48 (1.2)
Duration of principal recurrent pain ^b						
Short	193 (9.4)	199 (10.0)	392 (9.7)	157 (7.6)	200 (10.1)	357 (8.8)
Medium	138 (6.7)	181 (9.1)	319 (7.9)	108 (5.3)	167 (8.4)	275 (6.8)
Long	183 (8.9)	222 (11.2)	405 (10.0)	209 (10.2)	203 (10.2)	412 (10.2)
Frequency of principal recurrent pain ^c						
Low	197 (9.6)	205 (10.3)	402 (10.0)	175 (8.5)	184 (9.3)	359 (8.9)
Medium	226 (11.0)	269 (13.6)	495 (12.3)	193 (9.4)	234 (11.8)	427 (10.6)
High	100 (4.9)	139 (7.0)	239 (5.9)	116 (5.7)	160 (8.1)	276 (6.8)
Intensity of principal recurrent pain ^d						
Low	146 (7.1)	176 (8.9)	322 (8.0)	86 (4.2)	94 (4.7)	180 (4.5)
Medium	227 (11.1)	257 (13.0)	484 (12.0)	234 (11.4)	291 (14.7)	525 (13.0)
High	137 (6.7)	163 (8.2)	300 (7.4)	162 (7.9)	193 (9.7)	355 (8.8)
Psychosocial trigger or context of principal recurrent pain ^e						
None	335 (16.3)	371 (18.7)	706 (17.5)	312 (15.2)	353 (17.8)	665 (16.5)
At least one	188 (9.2)	242 (12.2)	430 (10.7)	172 (8.4)	225 (11.3)	397 (9.8)
Restrictions in daily living due to principal recurrent pain ^f						
None	266 (13.0)	297 (15.0)	563 (13.9)	269 (13.1)	305 (15.4)	574 (14.2)
At least one	250 (12.2)	307 (15.5)	557 (13.8)	211 (10.3)	273 (13.8)	484 (12.0)

Note: Percentages shown may not sum to 100 due to rounding or omission of missing values from the table.

^aSites were recoded from the adult respondent's selection of anatomical sites where the child felt pain during the three months prior to interview, from a list that included: head, back, ear, abdomen, pelvis, arm, leg, chest, throat, tooth, and other. Other sites, if any, could be specified by the respondent in an open field.

^bDuration was recoded as short (≤3 months), medium (4-12 months), or long (>12 months).

^cFrequency was recoded as low (at most once a month), medium (more than once a month, but at most once a week), or high (more than once a week).

^dIntensity was recoded from combinations of the responses to the FPS and VAS questions as follows: low (FPS 1-3 and VAS < 30), medium (FPS 1-3 and 30 ≤ VAS < 60, or FPS 4 and VAS < 60), or high (FPS 5-6 or VAS ≥ 60).

^eTrigger and context variables were recoded into two mutually exclusive categories: any psychosocial trigger or context (including close-ended options: anger/dispute, agitation/nervousness, school conditions/work, family conditions, and sadness/loneliness; as well as open-ended responses, eg stress/anxiety, peer pressure, tantrum, jealousy, attempt to obtain secondary gains) versus no psychosocial trigger or context reported (including somatic and normalising/situational conditions, such as diseases, dysfunctions, treatments, growth-related changes, activities of daily living, sports/physical activity-related trauma, leisure time activities, and weather conditions).

^fRestrictions in daily living queried were missing school, not being able to meet with friends, having trouble eating, having trouble sleeping, and not being able to engage in leisure time activities.

age 7 and 20.9% at age 10. The prevalence of recurrent pain remained stable: 28.1% at age 7 and 26.3% at age 10 [PR 0.93 (95% CI 0.87, 1.00)].

Among children with recurrent pain ($n = 1136$ at age 7 and 1062 at age 10), there was an increase in the prevalence of high severity across the duration, frequency, and intensity features: 35.7% to 38.8% for duration over 12 months [PR 1.09 (95% CI 0.98, 1.21)], 21.0% to 26.0% for pain recurring more than once a week [PR 1.24 (95% CI 1.06, 1.44)], and 26.4% to 33.4% for high intensity pain (FPS 5-6 or VAS ≥ 60) [PR 1.27 (95% CI 1.11, 1.44)] (Table 1). The prevalence of at least one resulting restriction in daily living decreased from 49.0% at age 7 to 45.6% at age 10 [PR 0.93 (95% CI 0.85, 1.02)]. A breakdown by type of restriction is presented in Table S3.

There was a gradient of increasing risk of highest severity at age 10 with increasing severity at age 7 across all features (Figure 2 and Table 2). When compared to children whose mothers reported no pain at age 7, the risk of multisite pain at age 10 was highest among children with multisite pain at age 7 [RR 2.18 (95% CI 1.90, 2.50)]. The relative risk of long duration at age 10 was also highest among children with that same attribute at age 7 [RR 3.65 (95% CI 2.89, 4.62)], and similar associations were found for frequency [RR 4.43 (95% CI 3.19, 6.15)] and intensity [RR 3.84 (95% CI 2.94, 5.01)]. Psychosocial triggers or contexts of first episodes at age 7 were strongly associated with the same type of trigger/context at age 10

[RR 3.06 (95% CI 2.44, 3.84)], but also with organic or circumstantial triggers/contexts. Daily-living restrictions at age 7 also tracked up to age 10 [RR 3.49 (95% CI 2.85, 4.27)]. Also, across all features, children in intermediate severity categories at age 7 were generally more likely to escalate to the highest severity category at age 10 when compared with those with no pain or non-recurrent pain.

The composition of the optimal 11 pain clusters obtained using k-medoids is described in Table 3. The majority of children ($n = 2348$, 59.4% of all children included in the cluster analysis) were grouped in one of five clusters with non-recurrent or no pain at both ages (clusters 1, 2, 7, 9, and 11). Clusters 3 and 4 ($n = 637$, 16.1%) included children with decreasing pain severity, that is recurrent pain at age 7 but not at age 10. Clusters 5 and 10 ($n = 565$, 14.3%) grouped children with pain of increasing severity from ages 7 to 10.

Clusters 6 and 8 ($n = 404$, 10.2%) included children with recurrent pain at both ages 7 and 10. Cluster 8 ($n = 177$, 4.5%) grouped children with particularly severe recurrent pain, as seen from their high probabilities of multisite pain (74.6% and 66.7% at ages 7 and 10, respectively), pain of medium/high frequency (78.6% and 83.6%), pain of medium/high intensity (79.3% and 93.2%), pain with any psychosocial trigger/context (59.3% and 61.0%), and pain resulting in daily-living restrictions (72.2% and 84.6%). Although severity in cluster 8 was the highest across all measures examined, the most distinctive features of this cluster were the high probabilities

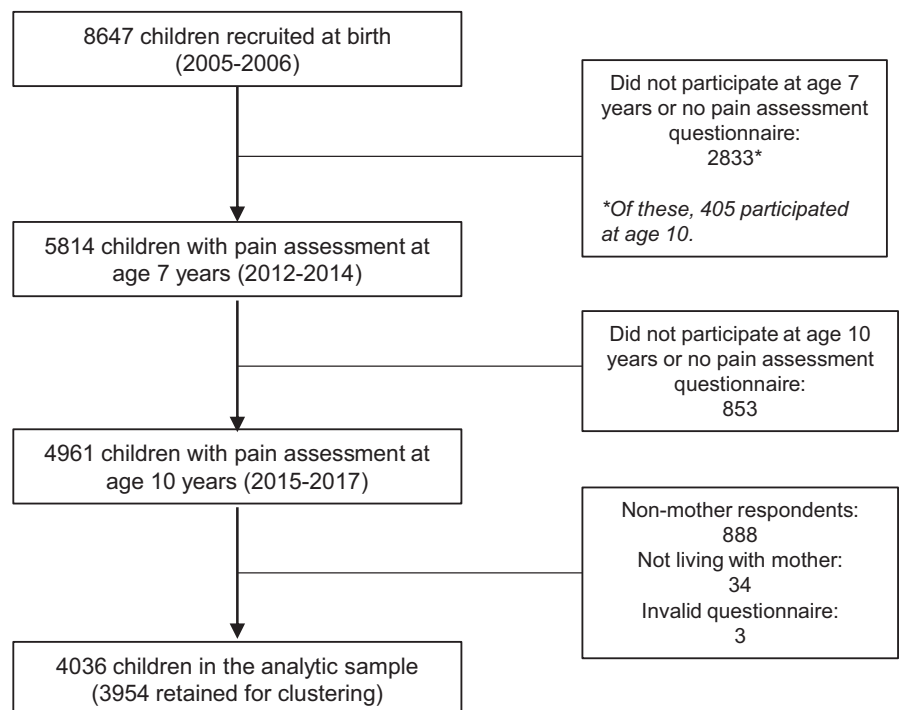


FIGURE 1 Selection of analytic sample from the Generation XXI birth cohort

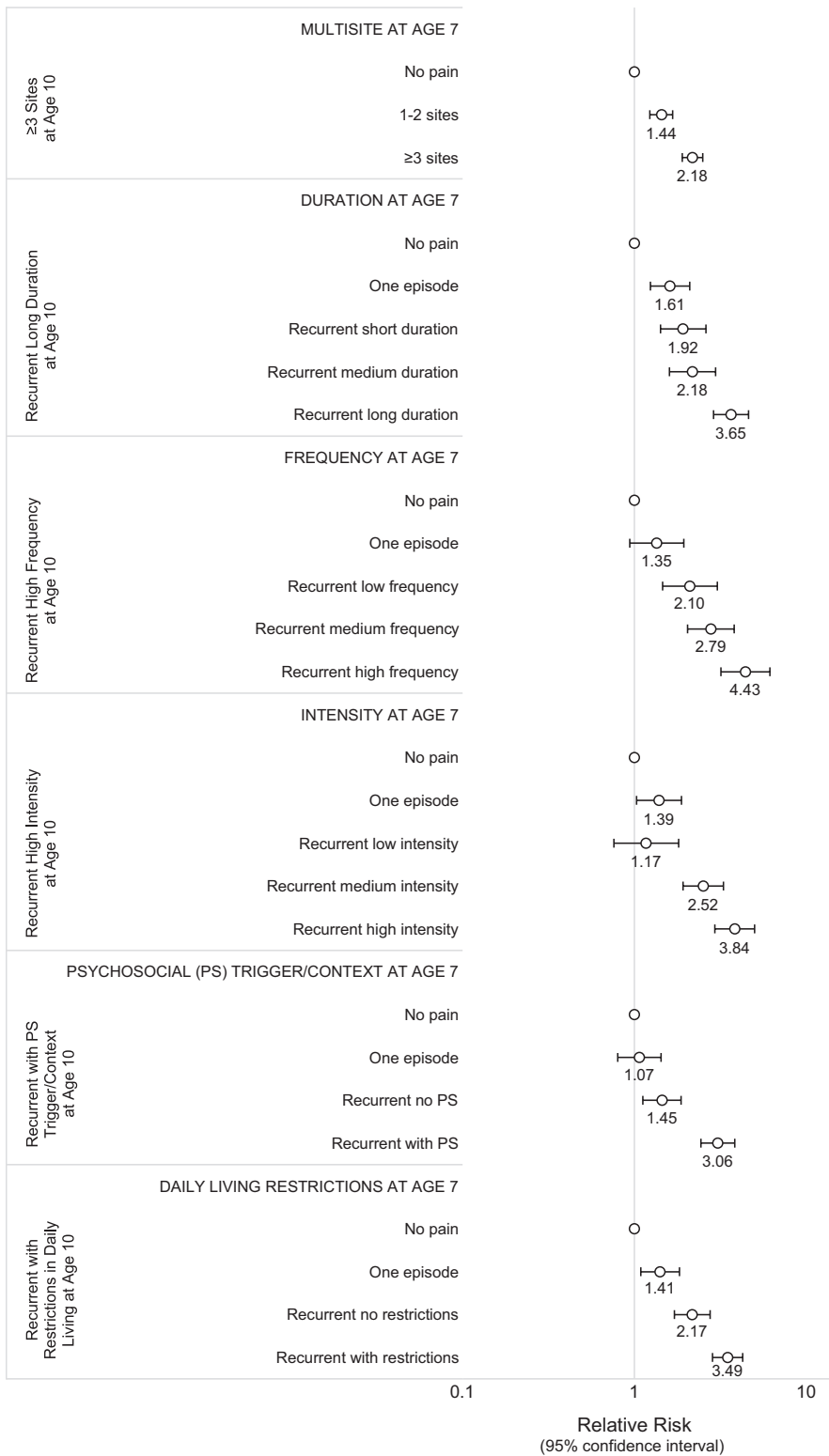


FIGURE 2 Relative risk and 95% confidence intervals for reporting pain of highest severity* at age 10 according to baseline reported severity for each pain feature at age 7. *Highest severity pain is characterised by mother reports of ≥ 3 pain sites, or of recurrent pain lasting more than 12 months, recurring more than once a week, with high intensity (as indicated by FPS ≥ 5 or VAS ≥ 60), any psychosocial trigger or context, or any resulting activity limitations

of multisite pain at age 7, daily-living restrictions at age 10, and psychosocial triggers/contexts in both evaluations. This is illustrated in Figure 3 by the distances between cluster 8 and its closest cluster in terms of the probabilities of each feature.

As for criterion validity of the cluster solution, the probability that mothers reported a family history of chronic pain was highest in cluster 8 (60.5%), followed by cluster 6 (48.5%), and under 20%

in the remaining clusters. Children in cluster 8 were themselves more likely to self-report any pain in the previous week at age 10 (58.3%), as well as pain affecting two or more anatomical regions (21.9%), and pain of higher intensity (self-reported FPS ≥ 4 ; 43.7%). Child-reported pain probability (any pain in the previous week) and severity (two or more pain sites and higher intensity) was intermediate among participants included in clusters 5, 6, and 10 (increasing

TABLE 2 Distribution, n, and conditional probabilities (%) of mother-reported pain features at age 10 given those reported at age 7 for pain episodes occurring in the three months prior to interview

Age 7	Age 10				
Multisite pain	No pain	1-2 sites	≥3 sites		
No pain	1067 (48.6)	791 (36.0)	339 (15.4)		
1-2 sites	333 (33.1)	451 (44.8)	223 (22.1)		
≥3 sites	217 (26.1)	335 (40.3)	280 (33.7)		
Duration ^a	No pain	One episode	Recurrent short duration	Recurrent medium duration	Recurrent long duration
No pain	1066 (49.3)	674 (31.2)	160 (7.4)	116 (5.4)	146 (6.8)
One episode	247 (36.3)	251 (36.9)	58 (8.5)	50 (7.4)	74 (10.9)
Recurrent short duration	114 (30.2)	132 (35.0)	50 (13.3)	32 (8.5)	49 (13.0)
Recurrent medium duration	92 (29.5)	96 (30.8)	46 (14.7)	32 (10.3)	46 (14.7)
Recurrent long duration	92 (23.7)	120 (30.8)	38 (9.8)	43 (11.1)	96 (24.7)
Frequency ^b	No pain	One episode	Recurrent low frequency	Recurrent medium frequency	Recurrent high frequency
No pain	1066 (49.2)	674 (31.1)	155 (7.2)	176 (8.1)	94 (4.3)
One episode	247 (36.2)	251 (36.8)	67 (9.8)	77 (11.3)	40 (5.9)
Recurrent low frequency	125 (31.7)	140 (35.5)	46 (11.7)	47 (11.9)	36 (9.1)
Recurrent medium frequency	115 (24.0)	145 (30.3)	66 (13.8)	95 (19.8)	58 (12.1)
Recurrent high frequency	63 (26.9)	69 (29.5)	25 (10.7)	32 (13.7)	45 (19.2)
Intensity ^c	No pain	One episode	Recurrent low intensity	Recurrent medium intensity	Recurrent high intensity
No pain	1066 (49.3)	674 (31.2)	81 (3.7)	212 (9.8)	130 (6.0)
One episode	247 (36.2)	251 (36.8)	37 (5.4)	90 (13.2)	57 (8.4)
Recurrent low intensity	92 (29.4)	103 (32.9)	29 (9.3)	67 (21.4)	22 (7.0)
Recurrent medium intensity	131 (27.9)	144 (30.7)	23 (4.9)	100 (21.3)	71 (15.1)
Recurrent high intensity	69 (23.4)	99 (33.6)	10 (3.4)	49 (16.6)	68 (23.1)
Psychosocial trigger or context ^d	No pain	One episode	Recurrent no psychosocial		Recurrent with psychosocial
No pain	1066 (49.2)	674 (31.1)	259 (12.0)		166 (7.7)
One episode	247 (36.2)	251 (36.8)	128 (18.8)		56 (8.2)
Recurrent no psychosocial	208 (30.4)	220 (32.1)	181 (26.4)		76 (11.1)
Recurrent with psychosocial	95 (22.5)	134 (31.8)	94 (22.3)		99 (23.5)
Restrictions in daily living ^e	No pain	One episode	Recurrent no limitation		Recurrent with limitations
No pain	1066 (49.4)	674 (31.2)	254 (11.8)		166 (7.7)
One episode	247 (36.3)	251 (36.9)	109 (16.0)		74 (10.9)
Recurrent no restrictions	164 (30.1)	166 (30.5)	124 (22.8)		91 (16.7)
Recurrent with restrictions	135 (25.0)	180 (33.3)	81 (15.0)		145 (26.8)

Note: Percentages shown may not sum to 100 due to rounding or omission of missing values in the table.

^aDuration of the child's recurrent principal pain is recoded as short (≤3 months), medium (4-12 months), or long (>12 months).

^bFrequency of the child's recurrent principal pain is recoded as low (at most once a month), medium (more than once a month, but at most once a week), or high (more than once a week).

^cIntensity of the child's recurrent principal pain is recoded from combinations of the responses to the FPS and VAS questions as follows: low (FPS 1-3 and VAS < 30), medium (FPS 1-3 and 30 ≤ VAS < 60, or FPS 4 and VAS < 60), or high (FPS 5-6 or VAS ≥ 60).

^dTrigger and context variables related to the child's recurrent principal pain are created based on elements from biopsychosocial and ecological models of disease causation and grouped into two mutually exclusive categories: any psychosocial trigger or context versus no psychosocial trigger or context reported.

^eRestrictions in daily living due to the child's recurrent principal pain were: missing school, not being able to meet with friends, having trouble eating, having trouble sleeping, and not being able to engage in leisure time activities.



TABLE 3 Cluster size and sex and feature distribution (%) for children (n = 3954) with mother-reported recurrent pain features at ages 7 and 10

	Cluster 1	Cluster 2	Cluster 3	Cluster 4	Cluster 5	Cluster 6	Cluster 7	Cluster 8	Cluster 9	Cluster 10	Cluster 11
Size (n)	257	292	342	295	391	227	730	177	476	174	593
% girls	44.0	41.1	61.4	46.1	56.3	42.7	44.7	72.3	100	63.2	0.0
Interpretation	No pain or non-recurrent	Non-recurrent	Decreasing severity	Decreasing severity	Increasing severity	Stable medium severity	No pain or non-recurrent	Stable higher severity	No pain	Increasing severity	No pain
Features at each age (%)	7 10	7 10	7 10	7 10	7 10	7 10	7 10	7 10	7 10	7 10	7 10
No pain	0.0 99.6	0.0 0.0	0.0 0.0	0.0 98.6	99.2 0.0	0.0 0.0	97.4 0.0	0.0 0.0	99.4 100	0.0 0.0	100 100
One episode	96.5 0.0	93.8 92.1	0.0 92.7	0.0 0.0	0.0 0.0	0.0 0.0	0.0 94.9	0.0 0.0	0.0 0.0	92.0 0.0	0.0 0.0
Multisite	29.6 0.4	29.5 24.0	44.2 28.7	47.5 1.4	0.5 38.4	54.6 32.6	2.6 25.5	74.6 66.7	0.6 0.0	40.8 58.6	0.0 0.0
Recurrent, medium/long duration	3.5 0.0	5.1 3.8	58.2 4.4	59.7 0.6	0.5 58.5	72.7 68.8	2.2 4.8	72.8 71.1	0.6 0.0	4.0 70.9	0.0 0.0
Recurrent, medium/high frequency	3.5 0.0	5.8 3.7	54.4 5.3	56.3 1.4	0.6 63.9	71.3 55.5	2.4 3.2	78.6 83.6	0.6 0.0	6.3 69.0	0.0 0.0
Recurrent, medium/high intensity	2.3 0.4	2.1 5.8	75.1 5.9	69.0 1.0	0.3 80.0	67.8 81.0	0.9 4.7	79.3 93.2	0.4 0.0	4.6 81.6	0.0 0.0
Recurrent, any psychosocial trigger	3.5 0.4	4.1 3.4	33.3 2.9	28.5 1.0	0.8 35.5	31.7 22.5	1.8 3.7	59.3 61.0	0.6 0.0	4.0 27.6	0.0 0.0
Recurrent, any restriction in daily living	3.5 0.4	1.0 4.5	56.5 2.9	42.3 1.4	0.8 34.2	33.2 33.0	0.8 4.8	72.2 84.6	0.6 0.0	4.0 36.4	0.0 0.0
Variables not used for clustering											
Family history of chronic pain for those with recurrent pain	0.6	1.0	16.8	12.3	16.8	48.5	1.3	60.5	0.1	18.4	0.0

(Continues)

TABLE 3 (Continued)

	Cluster 1	Cluster 2	Cluster 3	Cluster 4	Cluster 5	Cluster 6	Cluster 7	Cluster 8	Cluster 9	Cluster 10	Cluster 11
Doctor's visit due to recurrent pain	1.0	2.7	23.1	17.6	18.0	37.5	2.1	53.7	0.2	18.4	0.0
Child at age 10: any pain in previous week	24.8	40.2	38.2	19.5	49.2	49.5	42.0	58.3	20.2	41.8	22.6
Child at age 10: 2 + pain sites	5.7	9.6	9.5	4.5	12.0	16.3	10.3	21.9	7.0	9.6	5.9
Child at age 10: higher intensity pain	12.6	26.2	25.7	11.6	30.6	29.1	25.0	43.7	12.4	28.1	12.7

severity or stable medium severity, according to parents), followed by the remaining clusters (decreasing severity, non-recurrent, or no pain) (Table 3).

4 | COMMENT

4.1 | Principal findings

In this population-based birth cohort, mother-reported adverse pain experience tracked with a positive gradient between ages 7 and 10: the higher the severity of pain at baseline the higher the probability of reporting severe pain three years later. This happened across all pain features examined. Although most children had transient pain forms or no reported pain, a small cluster remained highly likely to have severe pain, with notably high probabilities of multisite pain with psychosocial attributions and daily-living restrictions.

4.2 | Strengths of the study

We explored a wide set of pain features collected as part of a large population-based prospective cohort study. Even though we did not attempt to identify sensitive periods, our finding of a measurable escalation of symptoms in the 7-10 age range may ultimately contribute a baseline scenario to design interventions for reshaping pain trajectories, as previously suggested.²³ The present findings are also essential from an exploratory standpoint, since they point to specific clusters and features of maternal-reported pain that may be particularly useful to track the course of pain into adolescence. The 13-year-old follow-up wave of the Generation XXI cohort includes self-reported pain history and will allow to test whether the present findings will indeed contribute to identify long-term adverse pain experiences into adolescence.

4.3 | Limitations of the data

Like most interval cohorts, the composition of the Generation XXI sample has been affected by nonparticipation. While differential losses may have affected the relative frequency of different pain forms, it is less likely that they would influence our findings on pain course within groups, as shown in our sensitivity analysis. Due to our population-based design, we assumed that the majority of pain reports originated in the absence of serious organic lesion, but we did not have a clinical validation. Of all children whose mothers reported recurrent pain at either evaluation, 40.6% had consulted a doctor due to that pain. This may have influenced the valuation of the child's pain experience. Also, pain report was obtained from the mother and we do not have information from fathers or other caregivers. As a result, our analyses likely overestimated the maternal contribution to the child's pain experience and may have induced some sex-related

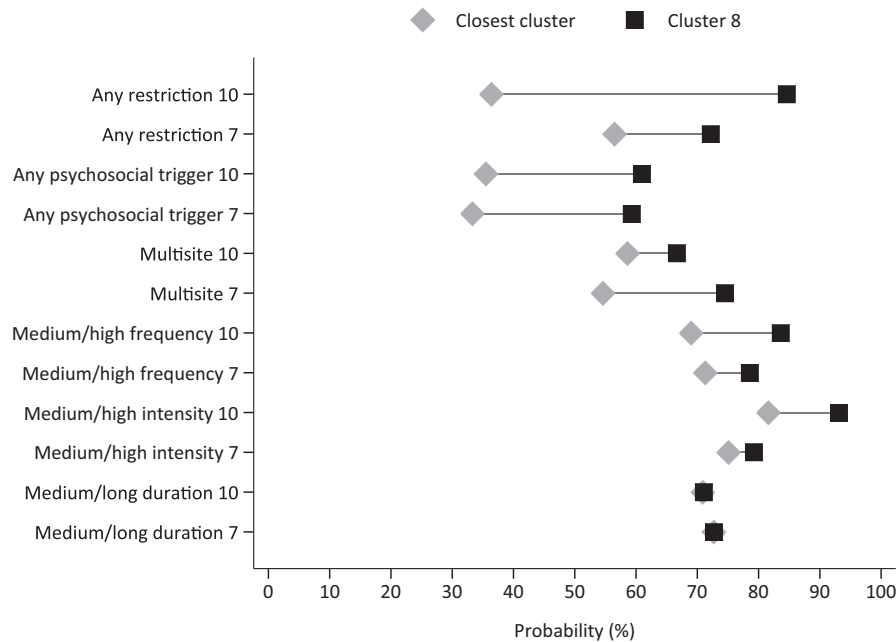


FIGURE 3 Equiplot of distances between the probabilities (%) of each feature at each age (7 and 10 years) in cluster 8 (black square) and in the cluster with the closest probability (grey diamond)*. *Closest clusters, in grey, differ by feature and are cluster 3 for medium/high intensity at age 7, any psychosocial trigger at age 7, and any restriction at age 7; cluster 5 for any psychosocial trigger at age 10; cluster 6 for multisite at age 7, medium/long duration at age 7, and medium/high frequency at age 7; cluster 10 for medium/long duration at age 10, medium/high frequency at age 10, medium/high intensity at age 10, multisite at age 10, and any restriction at age 10. The equiplot graphic representation was developed by the International Center for Equity in Health: equidade.org/equiplot

misclassification, as previously documented.²⁴ Finally, we had information on pain experience for only ages 7 and 10. Symptoms may have varied substantially between these moments, and we cannot also exclude the possibility that key aetiological periods occurred before age seven.

4.4 | Interpretation

The probability of maternal report of more severe pain at age 10 was highest among children in the highest severity category at age 7, in agreement with a documented stability of severe pain in childhood^{25,26} and into adulthood.²⁷ Also in line with previous literature,²⁸ the whole sample showed an increase in the prevalence of pain of higher severity across features. Our study adds that aggravation of pain was more frequent in children with non-recurrent pain or recurrent pain of lower severity when compared to those with no pain at baseline. This gradient argues for the relevance of intermediate severity for pain escalation.

In terms of the typology and relative frequency of major pain forms, our study is also in agreement with previous groupings in adolescents.⁷⁻⁹ We described 11 statistical clusters that use sex at birth and multiple pain features to improve discrimination. For comparison purposes, these can be summarised in four larger groups: five clusters grouping a majority of children without recurrent pain, two with decreasing pain between ages 7 and 10, two with increasing pain, and the remaining two clusters comprising those with recurrent pain in both evaluations. Our multi-feature approach allowed

to distinguish a group of children (cluster 8) with particularly adverse pain experiences, including a predominance of multisite pain, psychosocial triggers/contexts, and daily-living restrictions. These children also ranked highest in all child-reported variables on pain history and severity, which supports the validity of our approach. Interestingly, classical pain attributes such as duration and intensity were not particularly distinctive of this cluster.

Children with reported psychosocial triggers or contexts at age 7 were likely to be assigned similar attributions at age 10, and those triggers/contexts were very frequent in the highest severity cluster. Our interpretation is that causal attributions may be particularly relevant to identify early maladaptive pain. Attributional styles featuring psychological causes seem to determine clinical presentation as well as somatisation and psychologisation of distress in adults.¹⁰ Causal attributions in children also seem to play an important role in the whole pain experience,^{11,12,29} and their relevance for prognosis is illustrated by a lower likelihood of recurrence in children with traumatic pain aetiology than among those with non-traumatic pain.³⁰ Our results also support multisite pain at age 7 as a potential predictor of prognosis, regardless of specific topography, as shown in adults.³¹ We suggest that a brief classification of number of pain sites and causal attributions, which are not typically collected outside specialised paediatric pain clinics, may be useful in broader contexts to distinguish particularly adverse experiences in the absence of organic disease.

A central assumption of the present study relates to the value of maternal report. Interviewing mothers provided a comprehensive

description of pain at ages when children might be less able to quantify complex features. Maternal report results from the interaction between child-level factors (from nociception to pain behaviour) and parent-level factors, as well as contextual aspects. Parental affect, cognition, and behaviour towards pain—which we did not measure in this cohort—are consensual influences on children's pain experience^{32,33} and form a fundamental component of current models on the intergenerational transmission of chronic pain.^{4,34,35} In practice, maternal report can be seen simultaneously as an imperfect surrogate and a determinant of the child's pain experience, the latter being supported by our observation of an association between family history of chronic pain and child-reported pain. Empirical research supports a strong statistical association between parent- and self-reported child pain,^{36,37} although parents seem more likely to underestimate pain of lower severity.³⁸ From our brief assessment of the child's experience, maternal report seems to be valuable, since children assigned to the highest severity cluster were themselves more likely to report recent pain. External comparisons are particularly challenging due to heterogeneity in case definitions. A population-based investigation in the Netherlands with very similar methods to our study found a 23.7% prevalence of pain with duration over three months in children aged 8-11, which is remarkably close to our estimates.³⁹ The crude prevalence of multisite pain in our cohort was similar to the 20.6% pooled estimate recently obtained for 42 countries but higher than in the Portuguese subsample (13.8%).⁴⁰ In the same study, which included children aged 11-15, headache and backache were predominant with back pain being the most frequent complaint in Portugal, whereas in our sample abdominal pain was more frequent. Sex differences were also more pronounced than in our study. Even though we cannot exclude differences in sampling frames, participation, and data collection methods, age differences may explain this disparity since there is robust evidence of an increasing burden of headache and backache with increasing age, accompanied by stable or decreasing abdominal pain frequency, along with widening sex differences.²⁸

5 | CONCLUSIONS

In our study, all pain features tracked with a clear gradient between ages 7 and 10, arguing for the significance of the first decade of life in the escalation of the pain experience. Our finding of a cluster of children whose mothers reported multiple pain sites and psychosocial attributions since age 7 suggest that these may be distinctive early markers of a prolonged pain experience.

ACKNOWLEDGEMENTS

We thank the families enrolled in Generation XXI for their kindness, the members of the research team for their enthusiasm and perseverance, and the participating hospitals and their staff for their help and support. We also thank Ana Cristina Santos for her role in the coordination of the Generation XXI cohort, the field work team for their dedication, Sara Lourenço for her contribution in an early

stage of this work, and Sara Soares for her helpful suggestions on this manuscript.

ORCID

Raquel Lucas  <https://orcid.org/0000-0002-4408-8134>
 Makram Talih  <https://orcid.org/0000-0002-8834-5016>

TWITTER

Raquel Lucas  @rakalucas
 Henrique Barros  @hdpbarros

REFERENCES

1. Schechter NL, Palermo TM, Walco GA, Berde CB. Persistent pain in children. In: Bonica's management of pain. Editors: Fishman S, Ballantyne J, Rathmell JP, Bonica JJ. 4th ed. Baltimore, MD: Lippincott, Williams & Wilkins, 2010; pp. 767-782.
2. Dunn KM, Hestbaek L, Cassidy JD. Low back pain across the life course. *Best Pract Res Clin Rheumatol*. 2013;27:591-600.
3. Ben-Shlomo Y, Kuh D. A life course approach to chronic disease epidemiology: conceptual models, empirical challenges and interdisciplinary perspectives. *Int J Epidemiol*. 2002;31:285-293.
4. Stone AL, Wilson AC. Transmission of risk from parents with chronic pain to offspring: an integrative conceptual model. *Pain*. 2016;157:2628-2639.
5. Holley AL, Palermo TM. Introduction to the Special Issue: Advances in Behavioral and Psychological Pain Research in Children: From Prevention Through Chronic Pain Management. *J Pediatr Psychol*. 2018;43(3):219-223.
6. Krauss BS, Calligaris L, Green SM, Barbi E. Current concepts in management of pain in children in the emergency department. *Lancet*. 2016;387:83-92.
7. Dunn KM, Jordan KP, Mancl L, Drangsholt MT, Le Resche L. Trajectories of pain in adolescents: a prospective cohort study. *Pain*. 2011;152:66-73.
8. Coenen P, Smith A, Paananen M, O'Sullivan P, Beales D, Straker L. Trajectories of Low Back Pain From Adolescence to Young Adulthood. *Arthritis Care Res (Hoboken)*. 2017;69:403-412.
9. Isensee C, Hagmayer Y, Fernandez Castela C, Kroner-Herwig B. Paediatric headache trajectories - a reappraisal after nine years. *Cephalgia*. 2018;38:487-495.
10. Robbins JM, Kirmayer LJ. Attributions of common somatic symptoms. *Psychol Med*. 1991;21:1029-1045.
11. Crushell E, Rowland M, Doherty M, et al. Importance of parental conceptual model of illness in severe recurrent abdominal pain. *Pediatrics*. 2003;112:1368-1372.
12. Heathcote LC, Williams SE, Smith AM, Sieberg CB, Simons LE. Parent Attributions of Ambiguous Symptoms in Their Children: A Preliminary Measure Validation in Parents of Children with Chronic Pain. *Children (Basel)*. 2018;5(6):76.
13. Alves E, Correia S, Barros H, Azevedo A. Prevalence of self-reported cardiovascular risk factors in Portuguese women: a survey after delivery. *Int J Public Health*. 2012;57:837-847.
14. Larsen PS, Kamper-Jørgensen M, Adamson A, et al. Pregnancy and birth cohort resources in Europe: a large opportunity for aetiological child health research. *Paediatr Perinat Epidemiol*. 2013;27:393-414.
15. Roth-Isigkeit A, Thyen U, Stoven H, Schwarzenberger J, Schmucker P. Pain among children and adolescents: restrictions in daily living and triggering factors. *Pediatrics*. 2005;115:e152-162.
16. Bieri D, Reeve RA, Champion GD, Addicoat L, Ziegler JB. The Faces Pain Scale for the self-assessment of the severity of pain experienced by children: development, initial validation, and preliminary investigation for ratio scale properties. *Pain*. 1990;41:139-150.



17. Price DD, McGrath PA, Rafii A, Buckingham B. The validation of visual analogue scales as ratio scale measures for chronic and experimental pain. *Pain*. 1983;17:45-56.
18. Kaufman L, Rousseeuw PJ. *Finding Groups in Data : An Introduction to Cluster Analysis*. New York: Wiley; 1990.
19. Venables WN, Ripley BD. *Modern applied statistics with S [Chapter 11]*, 4th edn. New York: Springer; 2002.
20. Gower JC. General Coefficient of Similarity and Some of Its Properties. *Biometrics*. 1971;27:857-874.
21. Hennig C, Liao TF. How to find an appropriate clustering for mixed-type variables with application to socio-economic stratification. *J Royal Statist Soci Ser C-Appl Statist*. 2013;62:309-369.
22. R Core Team. R: A language and environment for statistical computing. Vienna, Austria: R Foundation for Statistical Computing; 2019. <https://www.R-project.org/>
23. Kongsted A, Kent P, Axen I, Downie AS, Dunn KM. What have we learned from ten years of trajectory research in low back pain? *BMC Musculoskelet Disord*. 2016;17:220.
24. Najman JM, Williams GM, Nikles J, et al. Bias influencing maternal reports of child behaviour and emotional state. *Soc Psychiatry Psychiatr Epidemiol*. 2001;36:186-194.
25. Kjaer P, Wedderkopp N, Korsholm L, Leboeuf-Yde C. Prevalence and tracking of back pain from childhood to adolescence. *BMC Musculoskelet Disord*. 2011;12:98.
26. Aartun E, Hartvigsen J, Wedderkopp N, Hestbaek L. Spinal pain in adolescents: prevalence, incidence, and course: a school-based two-year prospective cohort study in 1,300 Danes aged 11-13. *BMC Musculoskelet Disord*. 2014;15:187.
27. Brattberg G. Do pain problems in young school children persist into early adulthood? A 13-year follow-up. *Eur J Pain*. 2004;8:187-199.
28. King S, Chambers CT, Huguet A, et al. The epidemiology of chronic pain in children and adolescents revisited: a systematic review. *Pain*. 2011;152:2729-2738.
29. Claar RL, Walker LS. Maternal attributions for the causes and remedies of their children's abdominal pain. *J Pediatr Psychol*. 1999;24:345-354.
30. El-Metwally A, Salminen JJ, Auvinen A, Kautiainen H, Mikkelsen M. Lower limb pain in a preadolescent population: prognosis and risk factors for chronicity—a prospective 1- and 4-year follow-up study. *Pediatrics*. 2005;116:673-681.
31. Natvig B, Ihlebæk C, Kamaleri Y, Bruusgaard D. Number of pain sites – a simple measure of population risk? In: *Chronic pain epidemiology : from aetiology to public health*. Editors: Croft P, Blyth F, Windt Dvd. Oxford: Oxford University Press, 2010; pp. 71-79.
32. Ramchandani PG, Stein A, Hotopf M, Wiles NJ, Alspac Study T. Early parental and child predictors of recurrent abdominal pain at school age: results of a large population-based study. *J Am Acad Child Adolesc Psychiatry*. 2006;45:729-736.
33. Clementi MA, Faraji P, Poppert Cordts K, et al. Parent Factors are Associated With Pain and Activity Limitations in Youth With Acute Musculoskeletal Pain: A Cohort Study. *Clin J Pain*. 2019;35:222-228.
34. Palermo TM, Chambers CT. Parent and family factors in pediatric chronic pain and disability: an integrative approach. *Pain*. 2005;119:1-4.
35. Hoftun GB, Romundstad PR, Rygg M. Association of parental chronic pain with chronic pain in the adolescent and young adult: family linkage data from the HUNT Study. *JAMA Pediatr*. 2013;167:61-69.
36. Vetter TR, Bridgewater CL, Ascherman LI, Madan-Swain A, McGwin GL Jr. Patient versus parental perceptions about pain and disability in children and adolescents with a variety of chronic pain conditions. *Pain Res Manag*. 2014;19:7-14.
37. Lifland BE, Mangione-Smith R, Palermo TM, Rabbitts JA. Agreement Between Parent Proxy Report and Child Self-Report of Pain Intensity and Health-Related Quality of Life After Surgery. *Acad Pediatr*. 2018;18:376-383.
38. Sundblad GM, Saartok T, Engstrom LM. Child-parent agreement on reports of disease, injury and pain. *BMC Public Health*. 2006;6:276.
39. Perquin CW, Hazebroek-Kampschreur AAJM, Hunfeld JAM, et al. Pain in children and adolescents: a common experience. *Pain*. 2000;87:51-58.
40. Gobina I, Villberg J, Välimaa R, et al. Prevalence of self-reported chronic pain among adolescents: Evidence from 42 countries and regions. *Eur J Pain*. 2019;23:316-326.

SUPPORTING INFORMATION

Additional supporting information may be found online in the Supporting Information section.

How to cite this article: Lucas R, Talih M, Monjardino T, Guimarães S, Barros H. Mother-reported pain experience between ages 7 and 10: A prospective study in a population-based birth cohort. *Paediatr Perinat Epidemiol*. 2021;35:359-370. <https://doi.org/10.1111/ppe.12730>