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Offspring of Parents with Chronic Pain: A Systematic Review and Meta-Analysis of Pain, Health, Psychological, and Family Outcomes --Manuscript Draft--

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1 REVIEW & META-ANALYSIS OF PARENTAL CHRONIC PAIN (Higgins et al.)
2
34 **Abstract**
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55 *Keywords:* chronic pain, parents, offspring, children, systematic review, meta-analysis,
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Abstract

Offspring of parents with chronic pain may be at risk for poorer outcomes than offspring of healthy parents. The objective of this research was to provide a comprehensive mixed-methods, systematic synthesis of all available research on outcomes in offspring of parents with chronic pain. A systematic search was conducted for published articles in English examining pain, health, psychological, or family outcomes in offspring of parents with chronic pain. **Fifty-nine** eligible articles were identified (**31** population-based, **25 clinical**, 3 qualitative), including offspring from birth to adulthood and parents with varying chronic pain diagnoses (e.g., mixed pain samples, arthritis). Meta-analysis was used to synthesize the results from population-based and **clinical** studies, while meta-ethnography was used to synthesize the results of qualitative studies. Increased pain complaints were found in offspring of mothers and of fathers with chronic pain, and when both parents had chronic pain. Newborns of mothers with chronic pain were more likely to have adverse birth outcomes, including low birthweight, preterm delivery, caesarean section, intensive care admission, and mortality. Offspring of parents with chronic pain had greater externalizing and internalizing problems and poorer **social competence and** family outcomes. No significant differences were found on teacher-reported externalizing problems. The meta-ethnography identified six key concepts (developing independence, developing compassion, learning about health and coping, missing out, emotional health, and struggles communicating with parents). Across study designs, offspring of parents with chronic pain had poorer outcomes than other offspring, although the meta-ethnography noted some constructive impact of having a parent with chronic pain.

Keywords: chronic pain, parents, offspring, children, systematic review, meta-analysis, meta-ethnography

Offspring of Parents with Chronic Pain: A Systematic Review and Meta-Analysis of Pain, Health, Psychological, and Family Outcomes

Chronic pain tends to aggregate in families [37,72], and vulnerability to chronic pain is thought to be the result of complex interactions between both environment and genetics [37,46,50,77,91]. It is known that parental mental and physical health conditions generally put offspring at risk for poorer outcomes. For example, children of mothers with depression are more likely to experience depression and other mental health problems compared to children of mothers without depression [22,31,54,71], and children of parents with cancer have higher levels of anxiety, depression, and behaviour problems than children of parents without cancer [74]. Fewer studies have been conducted to examine the effects of parental chronic pain on their offspring.

Previous studies that have examined offspring of parents with chronic pain have studied the effects of parental chronic pain on the offspring's own pain complaints [37], other aspects of their physical health [51,53], their psychological health [44], and variables related to family relationships [75]. While several studies have found that offspring of parents with chronic pain have poorer pain [37], health [51], and psychological [44] outcomes than other offspring, other studies have found no between-group differences on pain complaints [43], and psychological and family outcomes [75]. Additionally, few studies have explored possible differences in the effects of maternal versus paternal chronic pain on offspring, although at least one study has found such differences [37], and other research has explored differences in mothers' and fathers' interactions with children during child pain [32,60]. These conflicting results highlight the need for a comprehensive review of the literature including studies conducted in various disciplines looking at a variety of outcomes in offspring of parents with chronic pain.

One prior narrative review described 18 studies of children of parents with chronic pain [82]. The review concluded that children and adolescents whose parents have chronic pain are at risk for more pain complaints (particularly those similar to their parents), and more internalizing and externalizing symptoms than children and adolescents of parents without chronic pain. However, the previous review [82] contained several critical limitations, including an overly narrow search strategy, lack of meta-analysis, and lack of inclusion of population-based and qualitative studies. It is important to conduct a mixed methods review of the literature on offspring of parents with chronic pain in order to better understand the conflicting results found in the literature. In particular, qualitative research may offer a different perspective on this topic, as participants may address topics that researchers had not previously considered. Better understanding the effects of parental chronic pain on offspring may identify opportunities to provide interventions to improve outcomes in offspring of parents with chronic pain.

The aim of the present study was to improve on previous work by conducting a rigorous mixed-methods systematic review, employing both meta-analysis and qualitative synthesis of the full range of empirical research (population-based, **clinical**, and qualitative studies) examining the relationship between parental chronic pain and pain, health, psychological, and family outcomes in their offspring.

Method

Search Strategy

The electronic databases searched included PubMed, PsycINFO, CINAHL, and EMBASE. PubMed was chosen over Web of Science as it has been shown to be an optimal biomedical database [30]. Databases were searched from inception to August 2014. The search strategy included combinations of terms for parent (e.g., parent, mother, father, caregiver,

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4 family), pain (e.g., pain, arthritis, fibromyalgia, headache, neuropathy), child (e.g., infant, child,
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6 adolescent, young adult), and association (e.g., risk, influence, predictor, association), and was
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8 developed in consultation with a librarian specializing in systematic searches. The particular pain
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10 conditions included in the search strategy were decided upon through consensus with expert
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12 adult and pediatric pain researchers and clinicians. **Medical Subject Headings** (MeSH) terms and
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14 other key terms were included where possible; text terms were searched in all databases for
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16 entries including these terms in the title or abstract. See Appendix A for the complete search
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18 strategy for each database. **Additional hand searching of previous relevant research was also**
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20 **conducted.**

26 **Eligibility Criteria**

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28 Eligible studies were empirical studies (population-based, **clinical**, or qualitative)
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30 examining the relationship between parental pain and offspring pain, health, psychological, or
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32 family outcomes. Studies had to include measures of offspring outcomes (e.g., not just measures
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34 of the parent's parenting abilities). Studies including offspring of any age from infancy to
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36 adulthood were eligible. Only published articles written in English were included, **given that**
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38 **research suggests that this does not significantly affect meta-analysis results [59].** Included
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40 studies had to have parent(s) as the identified pain patient(s) in their samples (rather than
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42 offspring).

48 **Study Selection and Data Extraction**

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50 Study selection and data extraction were completed following accepted standards for
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52 systematic review procedures [34,47,81]. The titles of all of the citations generated in the
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54 systematic search were reviewed for relevance by the first author (KH). For titles that were
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56 identified as relevant, abstracts were retrieved and reviewed to determine whether they met the
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4 review inclusion criteria. For any studies where there was uncertainty about exclusion, input was
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6 sought from a co-author (KB). Full articles identified for inclusion were retrieved and coded
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8 using data extraction sheets designed for the present review. Data extraction sheets were
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10 developed by the first author (KH) with consultation from co-authors. Characteristics of the
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12 parent and offspring samples and outcome measures were recorded, and data was extracted for
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14 all outcomes measured in each study by the first author (KH). A subset of **approximately** 20% of
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16 identified studies was coded by an additional coder and disagreements were solved with
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18 consensus. The outcomes measured in the population and **clinical** studies were grouped into **four**
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20 **core domains** (pain, health, psychological, family) and other outcomes to assist in the analysis of
21
22 the wide variety of outcomes measured across studies. For continuous variables, means and
23
24 standard deviations were extracted from each study; for dichotomous variables, odds ratios with
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26 95% confidence intervals or frequencies and group sizes were recorded.
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33 **Meta-Analysis**

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36 Meta-analyses were completed following accepted procedures for using this method
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38 [34,52]. Outcome data extracted from population and **clinical** studies were grouped by **domain**
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40 (pain, health, psychological, family, other). Within each **core domain**, similar constructs were
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42 grouped together for meta-analysis. **At least two studies measuring the same outcome were**
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44 **required in order to conduct a meta-analysis on the outcome. Studies were only combined in**
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46 **meta-analyses if their measurement of the outcome was similar enough that the combining of**
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48 **data in a meta-analysis would result in an interpretable construct** [34]. Psychological outcomes
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50 were grouped into internalizing problems (including scales measuring general internalizing
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52 symptoms and symptoms of depression, anxiety, and obsessive-compulsive disorders) and
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54 externalizing problems (including scales measuring general externalizing problems, child
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behaviour problems, **delinquency and hyperactivity**). Given the lack of agreement between measures completed by different informants in previous studies [1,67], outcomes were additionally grouped based on the source reporting on the measure (parent-, child-, and teacher-reported outcomes). Analyses were conducted using RevMan 5.3 software. Random effects analyses were used for all analyses, as this statistic is appropriate for meta-analyses in which the true effect size is thought to differ across studies [34]. This is the case in the present review given the variety of parental pain conditions studied and the variety of measures used for each outcome. In order to perform a meta-analysis for a particular outcome, at least two studies providing appropriate data on the outcome were required. Heterogeneity amongst studies was measured using the I^2 statistic, and was interpreted using the following recommended guidelines: 0%-40% indicates that heterogeneity might not be important, 30%-60% may represent moderate heterogeneity, 50%-90% may represent substantial heterogeneity, 75%-100% represents considerable heterogeneity [34].

Continuous outcomes were analyzed using standardized mean difference. This is considered an appropriate statistical test for meta-analyses in which outcomes are measured on different scales across studies and when comparing mean scores across groups which are not randomly assigned [34,52], which is the case in the present review. Data were entered in the analysis such that higher scores for each variable were indicative of greater levels of problems. Consequently, a positive standardized mean difference with both limits of the confidence interval being positive indicates that children in the parental chronic pain group had higher scores (poorer functioning) for the outcome compared to children of control parents. When the same outcome was rated by both the offspring's mother and father, data were pooled using the following

formulas: mean = $[(\text{mean1} * N1) + (\text{mean2} * N2)] / (N1 + N2)$ and pooled SD = square root of $[(SD1^2(N1-1) + SD2^2(N2-1)) / (N1 + N2 - 2)]$.

For dichotomous data, odds ratios were calculated using random effects generic inverse variance analyses. This type of analysis weights study effect estimates using the inverse variance of the effect estimate, which allows effect sizes to be combined across studies in which frequencies and group sizes were not reported [34]. Given that many studies reported odds ratios and 95% confidence intervals, but did not provide the frequency data necessary to calculate them, this analysis was considered most appropriate. In studies where odds ratios were not reported, frequency data were used to calculate odds ratios in RevMan to be included in the generic inverse variance analyses. **Subgroup analyses were conducted using a chi-square test in RevMan 5.3 software.**

Qualitative Synthesis

Qualitative studies identified in the systematic search were synthesized using meta-ethnographic techniques. Meta-ethnography is a well-accepted form of qualitative synthesis that involves translating the results of primary studies into one another, in order for overarching themes or concepts to be generated across studies [15,61,64]. This process involves seven steps: 1) Getting started (determining research questions); 2) Deciding what is relevant to the initial interest (determining the scope of the synthesis); 3) Reading the studies; 4) Determining how the studies are related; 5) Translating the studies into one another; 6) Synthesizing translations; and 7) Expressing the synthesis [61]. These steps were followed for the qualitative studies identified in the search. Each study was read three times and the key concepts identified in each study were recorded independently by two coders, with disagreements resolved through consensus. Key concepts were reviewed across studies to determine those that commonly occurred across

1 studies. Upon identification of these concepts, a table was created describing each concept in
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4 studies. Upon identification of these concepts, a table was created describing each concept in
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6 each primary study (“translating” studies into one another). Translations were synthesized by
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8 reading each study and concept definition and describing the relationship between the studies
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10 based on the concepts. The synthesis is expressed in the results section of the present review.
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12 These methods are consistent with suggested methods for using meta-ethnographic techniques
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14 [9,15,16].
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19 Results

20 Search Results

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22 The screening process is illustrated in a flow diagram in following the PRISMA model
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24 [58] in Figure 1. The systematic search identified 16,450 unique citations after removal of
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26 duplicates. Titles of all identified articles were screened for relevance to the topic, resulting in
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28 117 articles for possible inclusion. Abstracts of the 117 articles were reviewed for meeting the
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30 eligibility criteria. From this, 59 articles were identified for inclusion. Abstracts were excluded
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32 for several reasons, including not published articles (e.g., conference abstracts; $n = 19$), not
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34 examining parental pain ($n = 12$), focusing on offspring with chronic pain rather than parents
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36 with chronic pain ($n = 9$), not measuring offspring outcomes ($n = 8$), not empirical studies (e.g.,
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38 topical reviews; $n = 5$), not written in English ($n = 4$). Additionally, one study was excluded
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40 because it included case data from only one offspring of a parent with chronic pain. The 59
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42 studies identified for inclusion fell into three categories. Two categories utilized quantitative
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44 methodology: population-based studies (studies in which outcomes were measured quantitatively
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46 and a population-based sample was used; that is, participants were not specifically recruited to be
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48 part of a parental pain group or control group) and clinical studies (studies in which outcomes
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50 were measured quantitatively and a parental pain group was specifically recruited; a control
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group may or may not have been recruited). The third category of identified studies was qualitative studies (studies conducting qualitative analyses; e.g., grounded theory analysis). Thirty-one population-based studies, 25 clinical studies, and three qualitative studies were included in the review. Table 1 shows the studies included in the systematic review, along with the parental pain samples used and types of outcomes measured.

Population-based studies. Thirty population studies reported on a total of 4,626,806 offspring (sample size not reported in [38]). Fourteen (45.16%) examined the relationship between maternal chronic pain and offspring outcomes, while the remaining 17 studies (54.84%) examined pain in either parent in relation to offspring outcomes. Population studies examined mixed forms of parental chronic pain ($n = 13$, 41.94%) and parental arthritis ($n = 6$, 19.35%), headaches ($n = 6$, 19.35%), migraines ($n = 4$, 12.90%), and back pain ($n = 2$, 6.45%). The offspring examined varied in age across studies, including newborns (defined as ages 1-30 days; $n = 10$, 32.26%), infants (1-23 months; $n = 3$, 9.68%), preschoolers (ages 2-5 years; $n = 4$, 12.90%), children (ages 6-12 years; $n = 10$, 32.36%), adolescents (ages 13-18 years; $n = 13$, 41.94%), and adult offspring (> 18 years; $n = 5$, 16.13%).

Clinical studies. Twenty-three clinical studies reported on a total of 1,610 offspring of parents with chronic pain (sample size not reported in [66,76]). Eighteen (72.00%) included control groups, with a total of 1,305 offspring of parents without chronic pain being included. Seven (28.00%) clinical studies examined offspring of mothers with chronic pain while 17 (68.00%) examined offspring of either parent with chronic pain and one (4.00%) examined offspring of fathers with chronic pain. Similarly to the population studies, clinical studies ($n = 25$) included mixed parent pain samples ($n = 9$, 36.00%), as well as parent samples with arthritis ($n = 5$, 20.00%), fibromyalgia ($n = 3$, 12.00%), headaches ($n = 2$, 8.00%), irritable bowel

syndrome ($n = 2$, 8.00%), migraines ($n = 1$, 4.00%), inflammatory bowel disease ($n = 1$, 4.00%), back pain ($n = 1$, 4.00%), and temporomandibular pain ($n = 1$, 4.00%). These studies also included offspring of a range of ages, including newborns ($n = 1$, 4.00%), infants ($n = 3$, 12.00%), preschoolers ($n = 5$, 20.00%), children ($n = 16$, 64.00%), adolescents ($n = 14$, 56.00%), and adult offspring ($n = 7$, 28.00%). One study did not report on the age of included offspring [66].

Qualitative studies. Three qualitative articles describing two participant samples were identified in the systematic search. One study [29] interviewed 21 children (ages 6-12) of mothers with various chronic pain diagnoses regarding their understanding of their mothers' pain and health, their feelings about their mothers' pain, and their own health. The remaining two studies described the same sample of participants [83,84]. This sample comprised 30 young adults (18-21 years) with either a mother or father with a variety of chronic pain disorders. Participants were interviewed and asked to reflect on their experiences as an adolescent growing up with a parent with chronic pain, including the strategies they used to manage these experiences.

Meta-Analysis

Pain outcomes.

Population-based studies. Sixteen population studies [5,7,8,13,24,33,37,43,48,49,63,65,72,79,89,90] examined offspring pain outcomes, although not all provided data sufficient for inclusion in the meta-analysis. Meta-analysis of six studies [13,24,33,37,43,72] ($N = 9965$ offspring) providing appropriate data for meta-analysis found that offspring of mothers with chronic pain were more likely to report pain complaints compared to offspring of control mothers ($OR = 1.59$, 95% CI [1.37, 1.85], $Z = 6.00$, $p < 0.00001$, $I^2 = 44\%$).

The same pattern was revealed in offspring-reported pain complaints in **five** studies [13,24,33,37,43] ($N = 6453$) of offspring of fathers with chronic pain ($OR = 1.30$, 95% CI [1.08, 1.57], $Z = 2.77$, $p = 0.006$, $I^2 = 30\%$) and in **four** studies [13,24,37,43] ($N = 2380$) of offspring with both parents having chronic pain ($OR = 1.61$, 95% CI [1.29, 2.01], $Z = 4.22$, $p < .0001$, $I^2 = 0\%$). Additionally, a similar pattern was found in **five** studies [24,33,37,43,63] ($N = 7034$) that examined offspring with any one parent having chronic pain (regardless of sex) compared to control offspring ($OR = 1.59$, 95% CI [1.32, 1.92], $Z = 4.90$, $p < 0.00001$, $I^2 = 57\%$). **A test of subgroup differences revealed no significant differences in odds of pain complaints between offspring of mothers versus fathers with chronic pain ($X^2 = 2.60$, $p = 0.11$, $I^2 = 61.5\%$).**

The remaining population studies measuring offspring pain outcomes could not be combined for meta-analysis. One study examining mother-reported offspring pain complaints found that offspring of mothers with chronic daily headaches were more likely to have chronic daily headaches compared to offspring of mothers with no lifetime history of headaches [7]. The remaining studies used dissimilar parental pain or control definitions and could not be combined with the other studies [8,48,65], did not report associations between parental pain and offspring pain despite measuring these variables [5,49,90], or reported data in a format that could not be combined with the other meta-analyzed studies (e.g., regression coefficients that could not be combined with the OR data in the generic inverse variance analysis)[79,89].

Clinical studies. **Thirteen clinical** studies reported on various offspring pain outcomes that could not be appropriately grouped for meta-analysis. These studies found significant between-group differences in the outcomes they measured, including offspring-reported number of pain sites [26], teacher-reported presence of offspring pain complaints [27], parent-reported frequency of stomachaches or abdominal pain [41], number of tender points determined using

dolorimetry assessment [17], fibromyalgia diagnosis [70], self-reported presence of headaches [56], mean frequency of headaches [57], and pain-related responses to hypothetical scenarios [68]. Two studies [2,39] reported on pain outcome analyses in groups of relatives including offspring, but did not report the offspring results separately.

The remaining three clinical studies examined relationships between parental pain and offspring pain outcomes but did not include control groups of unrelated healthy parents and their offspring. One study [18] found a prevalence rate of fibromyalgia of 28% in offspring of parents with fibromyalgia (reported as higher than the prevalence in the general population), while another [45] found prevalence rates of irritable bowel syndrome of 12%-20% in offspring of parents with irritable bowel syndrome (reported as comparable with the general population prevalence). A third study [19] found that 16.67% of their sample of offspring of parents with chronic pain had pain complaints themselves.

Health outcomes.

Population-based studies. Eight population studies [11,12,20,21,51,62,87,88] examined birth outcomes in newborns of women with chronic pain conditions and provided appropriate data for meta-analysis. Results of these meta-analyses are shown in Table 2. Significant group differences were found for low birthweight, being small for gestational age, preterm delivery, caesarian section, perinatal mortality or stillbirth, and admission to neonatal intensive care unit (NICU), with newborns of mothers with chronic pain being more likely to experience these adverse birth outcomes. Significant group differences were not found for Apgar score below seven at five minutes after birth, congenital abnormalities, and instrument assisted delivery.

Four population studies examined other health outcomes that could not be appropriately combined for meta-analysis. Two studies found that children of parents with chronic pain had

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4 increased self-reported medication use for headache [3] and increased odds of emergency
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6 department visits and hospitalizations [53] compared to control children. Two other studies
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8 found no significant group differences when measuring author-defined newborn risk categories
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10 [4] and parent-reported use of over-the-counter analgesics by children [42].
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14 **Clinical studies.** Four clinical studies examined measures of offspring general health
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16 [26–28] and were combined within meta-analyses. Significant group differences were found for
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18 offspring-reported ($SMD = 0.79$, 95% CI [0.38, 1.19], $Z = 3.80$, $p = 0.0001$, $I^2 = 44%$; $N = 241$)
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20 [26–28] and parent-reported ($SMD = 0.51$, 95% CI [0.18, 0.84], $Z = 3.05$, $p = 0.002$, $I^2 = 0%$; N
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22 $= 153$) [25–27] general health scales, with offspring of parents with chronic pain having poorer
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24 scores on these measures. Two clinical studies [57,80] ($N = 132$) examined parent-reported
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26 questionnaire measures of somatization, and meta-analysis revealed no significant group
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28 differences ($SMD = 0.30$, 95% CI [-0.10, 0.69], $Z = 1.46$, $p = 0.15$, $I^2 = 18%$). Other clinical
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30 studies found that parental pain groups had offspring with lower birthweight [14], increased
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32 medication use for gastrointestinal symptoms [41], and more illnesses and injuries [66]
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34 compared to control groups, but found no differences regarding daily physical activity or sleep
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36 habits [80].
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42 43 **Psychological outcomes.**

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45 **Population-based studies.** Three population studies examined offspring psychological
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47 outcomes but could not be appropriately combined for meta-analysis. One study [44] found that
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49 adolescents with both parents having chronic pain had increased risk of experiencing anxiety and
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51 depressive symptoms than adolescents of control parents, while this was not seen in children
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53 with only one parent with chronic pain. They also found an increased risk of conduct problems in
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55 girls whose mothers had chronic pain. Another study [6] found that maternal headache status
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predicted clinical-range scores on parent-reported general behaviour measures in children without migraines, but not in children with migraines. A third study [38] did not report on offspring psychological outcomes for parental pain groups separately from other parental condition groups (e.g., cancer, mental health diagnoses).

Clinical studies. Fourteen clinical studies reported on offspring psychological outcomes. The combined results of seven studies [23,25–28,57,75] ($N = 502$) revealed that offspring of parents with chronic pain scored higher on parent-rated measures of externalizing problems compared to control offspring ($SMD = 0.58$, 95% CI [0.31, 0.85], $Z = 4.23$, $p < 0.0001$, $I^2 = 47\%$). However, two studies [23,27] ($N = 106$) measuring teacher-reported child externalizing problems revealed no difference between groups on this variable ($SMD = 0.63$, 95% CI [-0.12, 1.37], $Z = 1.64$, $p = 0.10$, $I^2 = 71\%$). Meta-analysis of five studies [26–28,57,75] ($N = 428$) found increased levels of parent-reported internalizing problems in offspring of parents with chronic pain ($SMD = 0.60$, 95% CI [0.39, 0.81], $Z = 5.62$, $p < 0.00001$, $I^2 = 7\%$). Five studies [25–28,35] ($N = 469$) found the same pattern of results for offspring-reported internalizing problems ($SMD = 1.13$, 95% CI [0.85, 1.41], $Z = 7.83$, $p < 0.00001$, $I^2 = 40\%$). Offspring-reported internalizing problems included measures of anxiety [25–27,35], depression [25,27,28,35], and obsessive compulsive disorder symptoms [35].

Six studies [23,25,27,28,57,80] ($N = 413$) examined parent-reported offspring social competence and meta-analysis revealed a significant difference between groups on this variable, with offspring of parents with chronic pain having poorer social abilities ($SMD = 0.47$, 95% CI [0.27, 0.67], $Z = 4.62$, $p = 0.00001$, $I^2 = 0\%$). Two studies [23,27] ($N = 106$) examining teacher-reported child social competence found a similar pattern of results ($SMD = 0.76$, 95% CI [0.23, 1.28], $Z = 2.83$, $p = 0.005$, $I^2 = 41\%$). One study reported that adolescents of parents with chronic

pain had lower self-esteem than control adolescents [35], while others measured offspring self-concept [69] and internalizing problems [19,92] without control groups.

Family outcomes.

Population-based studies. No population studies examined family outcomes.

Clinical studies. Three clinical studies [28,57,75] ($N = 232$) measured parent-reported family functioning and meta-analysis indicated that family functioning was poorer in families with parental chronic pain compared to healthy control families ($SMD = 0.47$, 95% CI [0.20, 0.74], $Z = 3.46$, $p = 0.0005$, $I^2 = 0\%$). One study did not provide sufficient data for meta-analysis [25].

Other Outcomes.

Population-based studies. No population studies examined offspring outcomes other than the pain, health, psychological, and family outcomes described above.

Clinical studies. Eight clinical studies examined offspring outcomes not falling into any of the other categories (pain, health, psychological, family). Two studies examined offspring days absent from school during one school year [23,25] ($N = 45$) and meta-analysis revealed no significant differences between groups ($SMD = 0.36$, 95% CI [-0.42, 1.14], $Z = 0.90$, $p = 0.37$, $I^2 = 34\%$). The outcomes measured in the remaining clinical studies were not sufficiently similar to be combined in meta-analyses. These studies examined physiological [80], school related (e.g., school activity participation) [35,41,68], and peer-relationship variables [36]. An additional study examined children's behaviours in response to their parent's pain (e.g., keeping distance, rubbing parent's neck) [76]. Two of these studies did not include control groups [41,76]; the remaining studies did not find group differences on these variables.

Qualitative Synthesis

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4 The three identified qualitative studies were synthesized using meta-ethnographic
5 techniques. One study [29] described a sample of 21 children ages 6-12 of mothers with various
6 chronic pain diagnoses, while the remaining two studies [83,84] described one sample of 30
7 young adults (ages 18-21) who reflected on their adolescence with either a mother or father
8 having various chronic pain diagnoses. A thorough literature review revealed no
9 recommendations for handling overlapping samples in meta-ethnography; thus, both samples
10 were included given that they presented analyses of separate data collected from the group of
11 participants. A summary of the key study characteristics and results of the meta-ethnography is
12 provided in Appendix B. Six key concepts regarding the effects of parental chronic pain were
13 identified across studies. The six concepts were: (1) development of children's independence at
14 an early age; (2) development of compassion or empathy for others; (3) learning about health and
15 coping; (4) missing out on normal activities or aspects of childhood or adolescence; (5) effects
16 on participants' emotional health; (6) struggles with communication.
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36 All three studies discussed the *development of children's independence at an early age*,
37 and this was viewed both positively and negatively by participants. Many mothers and children
38 in Evans and de Souza [29] reported that children were independent and caring, and while
39 mothers typically viewed this as positive, some children did not enjoy being left alone or
40 unsupervised. Participants in Umberger et al. [83] described hardships relating to developing
41 independence at a young age, and one of the "life lessons" that emerged from the study was
42 "growing up too fast is not a good thing". Some participants in Umberger et al. [84] used
43 independence from their parent as a strategy for coping with their parent's pain. Participants also
44 discussed the difficulty of having to take care of their parents physically and emotionally.
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4 The second key concept that emerged across studies was the *development of compassion*
5 *or empathy for others*. Children in Evans and de Souza [29] were reported to be caring and
6 helpful with household tasks, and most children reported enjoying being able to help their
7 mothers. In Umberger et al. [84], some young adults reported being able to empathize with their
8 parents as adolescents and consider the complexity of their pain problems. They also reported
9 learning the life lesson that “it is important to look beneath the surface” and being able to
10 empathize with others [83].
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21 The third key concept that emerged across studies was *learning about health and coping*.
22 Children learned about health, illness, their own bodies, and strategies for coping with stress
23 because of their mother’s pain [29]. Many young adults in Umberger et al. [83] reported the
24 positive experience of having learned “life lessons” as adolescents because of growing up with a
25 parent with chronic pain. Learning about health and coping did not emerge as a key concept from
26 Umberger et al. [84].
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36 The fourth key concept was *missing out* on normal activities or aspects of childhood and
37 adolescence because of living with a parent with chronic pain. Children reported missing out on
38 social activities because of their mother’s pain, and some expressed anger because of this [29].
39 Some young adults reported intense feelings of grief as adolescents regarding the loss of their
40 normal childhood and questioned whether their parents loved them [84], while some learned to
41 accept missing out on certain aspects of life because of parental chronic pain [83].
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50 The fifth key concept that emerged was *effects on participants’ emotional health*.
51 Participants in all samples indicated that they had suffered emotionally at times due to the
52 experience of living with parental chronic pain. Children were described as “clingy” and
53 showing anxiety and sadness related to parental pain [29]. Young adults reported experiencing
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4 negative emotions as adolescents including sorrow, anger, frustration, and fear, both because of
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6 the difficulties of having a parent with chronic pain and the worry that their parent would not be
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8 around for important future events [84]. Some young adults also described worrying during their
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10 adolescence about whether their parent's pain was life threatening and whether they would
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12 experience chronic pain themselves in the future, as well as experiencing guilt when they
13
14 questioned whether their parents were truly in as much pain as they described [83].
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19 The final key concept that emerged was *struggles with communication*. Across studies,
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21 children tended to hide their true thoughts and feelings from their parent with pain and avoided
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23 communicating their needs. In Evans & de Souza [29], several children reported anxiety about
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25 their mother's pain, of which their mother was not aware. Some young adults reported distancing
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27 themselves from their parents both physically and emotionally in their adolescence, and did not
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29 discuss their feelings regarding their parent's pain with anyone [84]. However, those who were
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31 able to share their feelings with their parents as adolescents experienced an increased closeness
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33 in the relationship [84]. Some participants explained that they avoided initiating conversations
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35 with their parent about pain for fear that it would cause their parent additional pain and suffering
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37 [83]. Few participants across studies described receiving clear information about their parent's
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39 pain.
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45 One main difference in results emerged between the three qualitative studies; namely, the
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47 effects of parental chronic pain on offspring pain experiences. Evans and de Souza [29] reported
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49 that one of the most profound effects of maternal chronic pain was on the children's physical
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51 health, as many children reported experiencing pain complaints and discussed the positive
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53 aspects of having pain (e.g., receiving special attention from parents). This concept was not
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55 described in either of the other two included qualitative studies examining the young adult
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sample. While some participants in Umberger et al. [83] reported worrying about whether or not they would experience chronic pain problems in the future, they did not discuss effects of their parents' pain on their own pain or other aspects of physical health.

Discussion

This comprehensive and rigorous mixed-methods systematic review and syntheses indicates that overall, offspring of parents with chronic pain have poorer outcomes in the areas of pain, health, psychological, and family functioning as compared to offspring of parents without pain. Thirty-one population-based studies, 25 clinical studies, and three qualitative studies were included, representing parents with a variety of pain conditions and offspring from newborns to adulthood. Population studies indicated that offspring of parents with chronic pain were more likely to have pain complaints than other offspring, including when either one or both parents had chronic pain. No differences were found for maternal versus paternal chronic pain. They also indicated that newborns of mothers with chronic pain had increased rates of many, but not all, adverse birth outcomes, including low birthweight, small for gestational age, preterm birth, caesarian section, perinatal mortality, and admission to neonatal intensive care.

Clinical studies found that offspring of parents with chronic pain had poorer scores on general health measures, psychological outcomes, and family functioning than other offspring, including higher levels of parent-reported externalizing and internalizing problems and parent- and teacher-reported social competence. Differences were not found for teacher-reported externalizing problems, parent-reported somatization, or days absent from school.

Synthesis of the included qualitative studies also revealed negative effects of parental chronic pain on offspring, including a sense of missing out on a normal childhood or adolescence, negative effects on emotional health, and struggles with parent-child

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4 communication. However, the qualitative synthesis also uniquely identified positive outcomes
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6 associated with having a parent with chronic pain, including developing independence,
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8 developing compassion or empathy for others, and learning about health and coping. This may
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10 have revealed positive outcomes associated with having a parent with chronic pain because
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12 qualitative studies were designed in such a way that participants could discuss any of their
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14 experiences related to parental chronic pain, whether positive or negative, **while the quantitative**
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16 **studies (population and clinical)** focused on outcomes that were hypothesized to be poorer in
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18 children of parents with chronic pain.
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24 This review synthesized findings regarding the relationship between parental pain and
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26 offspring outcomes. A relationship between maternal chronic pain and several adverse birth
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28 outcomes was found, and the meta-analysis of outcomes based on the informant assessing the
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30 outcome (e.g., parent, offspring, teacher) highlighted important findings. Offspring of parents
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32 with chronic pain were found to have greater parent-reported externalizing problems, but not
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34 teacher-reported externalizing problems. This suggests that future studies should take into
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36 account different informants when studying the effects of parental chronic pain on offspring.
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38 This review also synthesized results that are generally consistent with the previous narrative
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40 review completed in this area [82]; offspring of parents with chronic pain were more likely to
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42 have pain complaints and internalizing and externalizing problems compared to other offspring.
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48 While it has been established that offspring of parents with chronic pain have poorer
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50 outcomes than offspring of healthy parents, it is unclear whether this finding is directly related to
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52 parental pain per se, or to other characteristics commonly associated with chronic pain, such as
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54 comorbid mental health issues, medication use, and/or pain-related disability. For example,
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56 parental depression is associated with poorer outcomes in offspring [22,31,54,71] and the
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4 prevalence of major depressive disorder is higher among adults with chronic pain than in the
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6 general population [55]. Two studies included in our review [35,36] controlled for parental
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8 mental health by including a comparison group of parents with depression in addition to chronic
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10 pain. These studies reported poorer outcomes for offspring of parents in both clinical groups
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12 compared to healthy control groups, but were inconsistent in observed differences between
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14 clinical groups. Two other studies found mixed results regarding correlations between parental
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16 mental health and child outcomes [57,66]. Similarly, medication use in mothers could have
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18 influenced birth outcomes in newborns, although this was not controlled for in any of the
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20 included population studies. Moreover, studies included in our review focused primarily on the
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22 impact of the presence of parental chronic pain on offspring outcomes. However, individuals
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24 with chronic pain can have highly variable levels of associated pain-related disability and
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26 interference [73,85] and therefore the impact of level of impairment as a result of pain on
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28 offspring outcomes will be an important focus for future work. Only two studies in our review
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30 included parental pain-related disability in their analyses of offspring outcomes [75,80].
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32 Attempts are needed to disentangle the unique contribution of parental chronic pain relative to
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34 other associated parental factors in influencing outcomes in offspring.
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43 The present systematic review, meta-analysis, and qualitative synthesis has many
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45 strengths. Firstly, a broad search strategy was used, which was successful in identifying a large
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47 number of relevant articles including a variety of study designs and samples (population, clinical,
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49 and qualitative studies). The inclusion of population studies is important because these studies
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51 are more representative of the general population of families dealing with chronic pain than
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53 matched control group studies. Completing a synthesis of qualitative studies allowed for the
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55 incorporation of the rich data gained in qualitative research in the review and for the discussion
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4 about both positive and negative self-perceived effects of having a parent with chronic pain.
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6 Secondly, this review is unique in its inclusion of both meta-analysis and meta-ethnographic
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8 techniques. Completing formal syntheses allowed for conclusions to be drawn from
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10 combinations of studies that would not have been possible in a narrative synthesis. In particular,
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12 the meta-analysis allows for estimates of the effect sizes of the studied relationships to be
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14 calculated. The use of a mixed methods approach to knowledge synthesis is innovative in the
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16 field of pain research and provides a richer basis for understanding the state of the research in
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18 this topic area.
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24 A limitation of this work is that an evaluation of the quality of the primary studies
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26 included in the review was not possible given that existing quality assessment tools could not be
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28 applied across the different types of studies (e.g., Grading of Recommendations Assessment,
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30 Development, and Evaluation (GRADE) criteria [10,40]). Furthermore, application of the
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32 GRADE criteria require that prognostic studies must use longitudinal designs, and only two of
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34 31 population studies [5,65] and two of 25 clinical studies [66,80] employed longitudinal
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36 designs. The meta-ethnographic synthesis of qualitative studies may have been limited by the
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38 small number of included studies. Furthermore, several of the quantitative meta-analyses
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40 included small numbers of studies.
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46 This review addresses an important knowledge synthesis gap and identifies areas in need
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48 of further exploration in the study of offspring of parents with chronic pain. Future studies
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50 should use prospective, longitudinal designs to examine parental pain as a prognostic factor in
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52 the development of chronic pain in offspring and to further examine the processes by which
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54 parental pain may transfer to offspring. While prior studies have examined other prognostic
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56 factors in pain development [86], few studies have used longitudinal designs to examine the role
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of parental chronic pain in offspring outcomes. Additionally, future studies should include multiple measures of outcomes and multiple informants, as relying only on parent reports of offspring pain may be biased by the parents' own pain experiences. Another gap in this area is the role of particular aspects of parental pain such as specific pain parameters (e.g., pain frequency, onset, and duration), pain-related disability, and parent mental health in the relationship between parental pain and offspring outcomes. No studies have examined the timing of onset of parent pain with respect to offspring age or developmental stage. **While no differences in outcomes of offspring of mothers versus fathers with chronic pain were found in this review, few included studies provided data separated by parent sex for meta-analysis. Future studies should examine possible differences in this area, given that at least one well-powered study has found such a difference [37].**

Thus far, population-based and **clinical** studies have established that in general, offspring of parents with chronic pain tend to have poorer outcomes in terms of pain, health, psychological, and family functioning as compared to offspring of parents without pain. However, as revealed in the qualitative synthesis, there are at least some perceived positive offspring outcomes of having a parent with chronic pain. Future research should move beyond simply comparing offspring with and without parental chronic pain, instead, focusing on possible predictors of *which* offspring of parents with chronic pain are at greatest risk for poorer pain, health, psychological, and family outcomes. Employing a risk-resilience framework [78] to understanding possible offspring, parent, dyadic, and family variables involved in predicting poorer and, conversely, better offspring outcomes will be important. A greater focus on the mechanisms involved in the relationship between parental chronic pain and offspring outcomes is also needed (i.e., *why* and *how* do these differences occur). **Researchers should also consider**

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4 the strengths of qualitative methodology when designing future studies, and consider whether
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6 this methodology may be appropriate for answering their particular research questions. While
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8 additional research is needed, the results of this review highlight the impact of parental chronic
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10 pain on offspring outcomes and the need for clinicians to incorporate this factor in routine
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12 chronic pain assessment and management.
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Figure Legends

Figure 1. PRISMA flow diagram of identification and screening process for systematic review.

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1 REVIEW & META-ANALYSIS OF PARENTAL CHRONIC PAIN

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3
4 **Summary**

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6 This review and synthesis concludes that offspring of parents with chronic pain have poorer pain,
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8 health, psychological, and family outcomes than offspring of healthy parents.
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Figure 1 Revised

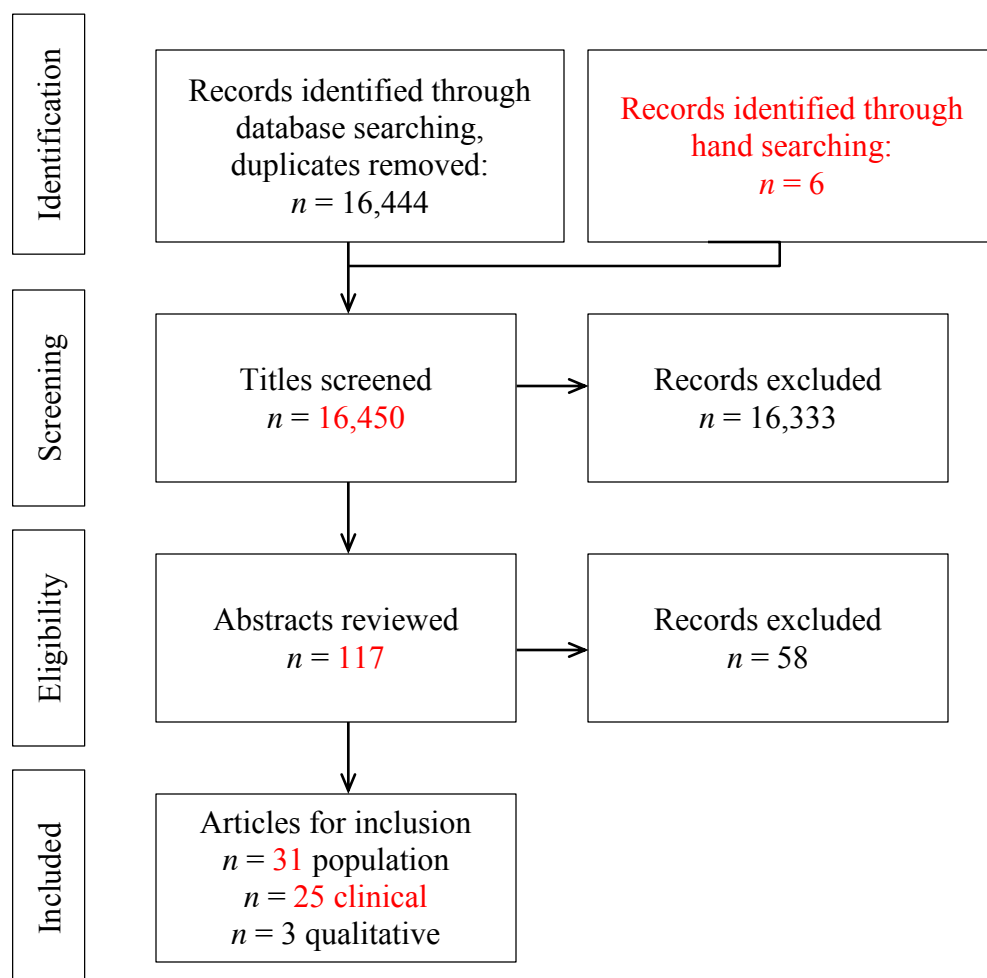


Table 1.

Characteristics of Studies Included in the Systematic Review

First Author & Year	Parent with Pain	Type of Parental Pain	Offspring Age Range	Number of Offspring	Offspring Outcomes Measured				
					Pain	Health	Psych	Family	Other
<i>Population</i>									
Andersen 2012 [3]	Either	Headaches	2-17 years	8256	x				
Aromaa 1996 [4]	Mother	Headaches	Newborn	1443	x				
Aromaa 1999 [5]	Either	Headaches	6 years	908	x				
*Arruda 2010 [7]	Mother	Headaches	5-12 years	1994	x				
Arruda 2012 [6]	Mother	Headaches	5-12 years	1856			x		
Assadi 2013 [8]	Either	Headaches	14-18 years	272	x				
*Banhidy 2007 [11]	Mother	Migraine	Newborn	38151		x			
*Blair 2011 [12]	Mother	Migraine	Newborn	660		x			
*Borge 2000 [13]	Either	Mixed	13-15 years	229	x				
*Chen 2010 [20]	Mother	Migraine	Newborn	29466		x			

*Chen 2013 [21]	Mother	Arthritis	Newborn	941574	x	
*Deubner 1977 [24]	Either	Migraine	10-20 years	232	x	
*Hasvold 1996 [33]	Either	Mixed	20-70 years	1939	x	
*Hoftun 2013 [37]	Either	Mixed	13-18 years	5370	x	
Holmes 2003 [38]	Either	Arthritis	> 18 years	NR		x
Jensen 2014 [42]	Mother	Mixed	6-11 years	131	x	
*Jones 2004 [43]	Either	Mixed	12-15 years	1293	x	
Kaasboll 2012 [44]	Either	Mixed	13-18 years	3227		x
Koutantji 1998 [48]	Either	Mixed	18-51 years	180	x	
Kovacs 2003 [49]	Either	Back pain	13-15 years	7361	x	
*Lin 2010 [51]	Mother	Arthritis	Newborn	11472	x	
Logan 2008 [53]	Either	Mixed	0-17 years	258313	x	
*Norgaard 2010 [62]	Mother	Arthritis	Newborn	871579	x	
*O'Sullivan 2008 [63]	Either	Back pain	14 years	1608	x	
Ramchandani 2006 [65]	Either	Mixed	6 years	8272	x	
*Saunders 2007 [72]	Mother	Mixed	11-17 years	2466	x	

Thomas 1992 [79]	Either	Mixed	> 18 years		141	x				
*Wallenius 2011 [88]	Mother	Arthritis	Newborn		1800827		x			
*Wallenius 2014 [87]	Mother	Arthritis	Newborn		627138		x			
Wilson 2014 [89]	Either	Mixed	11-14 years		173	x				
Wolff 2009 [90]	Mother	Mixed	14 months		275	x				
<i>Clinical</i>										
Aguas 2011 [2]	Either	IBD	> 18 years	$n_p = 74$		x				
				$n_p = 133,$						
Bowden 2001 [14]	Mother	Arthritis	0-8 months	$n_c = 103$			x			
Buskila 1996 [18]	Mother	FM	5-46 years	$n_p = 58$		x				
				$n_p = 67,$						
Buskila 1997 [17]	Mother	FM	> 8 years	$n_c = 310$		x				
Chaturvedi 1988 [19]	Either	Mixed	10-15 years	$n_p = 36$		x		x		
				$n_p = 35,$						
*Chun 1993 [23]	Either	Mixed	6-16 years	$n_c = 29$				x		x
*Dura 1988 [25]	Mother	Mixed	7-13 years	$n_p = 7,$			x	x	x	x

				$n_c = 14$				
				$n_p = 24,$				
*Evans 2007 [26]	Either	Mixed	6-12 years	$n_c = 12$	x	x	x	
				$n_p = 55,$				
*Evans 2007 [27]	Mother	Mixed	6-12 years	$n_c = 48$	x	x	x	
				$n_p = 55,$				
*Evans 2006 [28]	Mother	Mixed	6-12 years	$n_c = 48$		x	x	x
				$n_p = 16,$				
*Hirsch 1985 [35]	Either	Arthritis	12-18 years	$n_c = 16$			x	x
				$n_p = 16,$				
Hirsch 1985 [36]	Either	Arthritis	12-18 years	$n_c = 16$				x
				$n_p = 109,$				
Hudson 2004 [39]	Either	FM	> 18 years	$n_c = 53$	x		x	
				$n_p = 42,$				
Jamison 1992 [41]	Either	Mixed	6-18 years	$n_c = 55$	x	x		x
Kalantar 2003 [45]	Either	IBS	> 18 years	$n_p = 97$	x			

Messinger 1991 [56]	Either	Headaches	> 18 years	$n_p = 247,$ $n_c = 279$	x				
*Mikail 1990 [57]	Either	Mixed	9-17 years	$n_p = 24,$ $n_c = 30$	x	x	x	x	
Raphael 1990 [66]	Mother	TMPDS	NR	$n_p = NR,$ $n_c = NR$		x			
Rickard 1988 [68]	Father	Back pain	8-12 years	$n_p = 21$	x		x		x
Roy 1994 [69]	Either	Mixed	5-18 years	$n_p = 31$ $n_p = 357,$			x		
Saito 2010 [70]	Either	IBS	18-70 years	$n_c = 195$	x				
Smith 1998 [76]	Either	Migraine	0-17 years	$n_p = 52,$	NR				x
*Smith 2006 [75]	Either	Headaches	8-15 years	$n_c = 23$ $n_p = 25,$			x	x	
*Turner Cobb 1998 [80]	Either	Arthritis	4-16 years	$n_c = 53$		x	x		x

Zelkowitz 2013 [92]	Either	Arthritis	1.5-18 years	$n_p = 29$	x
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Qualitative

*Evans 2008 [29]	Mother	Mixed	6-12 years	$n_p = 21$
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*Umberger 2013 [84]	Either	Mixed	18-21 years	$n_p = 30$
*Umberger 2014 [83]				

Note. * indicates that the study was included in the meta-analyses or meta-ethnography. IBD = inflammatory bowel disease; FM = fibromyalgia; IBS = irritable bowel syndrome; NR = not reported; n_p = number of offspring in parental pain group; n_c = number of offspring in control group; **TMPDS = temporomandibular pain and dysfunction syndrome.**

Table 2

Results of Meta-Analyses of Newborn Birth Outcomes

Outcome	Studies	Number of Participants	OR	95% CI	I^2
Low birthweight	5	672408	1.35	[1.13, 1.60]	59%
Small for gestational age	6	2447410	1.24	[1.08, 1.42]	63%
Preterm delivery	8	2486221	1.59	[1.28, 1.97]	83%
C-section	6	2447410	1.51	[1.25, 1.83]	90%
Apgar score below 7 at 5 minutes	3	2071095	1.16	[0.82, 1.62]	0%
Perinatal mortality/stillbirth	3	1464898	2.03	[1.29, 3.20]	0%
NICU admission	3	1534893	1.69	[1.01, 2.81]	75%
Congenital abnormalities	3	1464898	1.22	[0.91, 1.64]	18%
Instrument assisted delivery	3	1534893	1.55	[0.82, 2.92]	88%

Note. OR = odds ratio; CI = confidence interval

Supplementary Materials - Appendix A

Search Strategy by Database

Table A1. Search Terms: PubMed

Parental Terms	Pain Terms	Child Terms	Relationship Terms
Title/abstract:	Title/abstract:	Title/abstract:	Title/abstract:
Parent* Mother* Maternal Father* Paternal Family Families Caregiver* Care-giver*	Pain* Fibromyalgia Irritable bowel syndrome Arthrit* Osteoarthrit* Headache* Migraine* Neuralgi* Neuropath* Complex regional pain syndrome	Child* Adolescen* Teen* Youth* Infant* Young adult*	Impact* Influenc* Predict* Factor* Association* Risk* Relation* Correlat* Aggregat*
MeSH Terms:	MeSH Terms:	MeSH Terms:	MeSH Terms:
Parents (Exp)	Pain (Not Exp) Chronic Pain (Exp) Fibromyalgia (Not Exp) Irritable Bowel Syndrome (Not Exp) Arthritis (Not Exp) Osteoarthritis (Not Exp) Headache (Not Exp) Migraine (Not Exp) Neuralgia (Not Exp) Peripheral Nervous System Diseases (Not Exp) Complex Regional Pain Syndromes (Not Exp)	Child (Exp) Child, Preschool (Exp) Adolescent (Exp) Infant (Exp) Young adult (Exp)	Risk (Not Exp) Association (Not Exp)

Note. Exp = exploding

Table A2. Search Terms: CINAHL

Parental Terms	Pain Terms	Child Terms	Relationship Terms
Title/abstract:	Title/abstract:	Title/abstract:	Title/abstract:
Parent* Mother* Maternal Father* Paternal Family Families Caregiver* Care-giver*	Pain* Fibromyalgia Irritable bowel syndrome Arthrit* Osteoarthrit* Headache* Migraine* Neuralgi* Neuropath* Complex regional pain syndrome	Child* Adolescen* Teen* Youth* Infant* Young adult*	Impact* Influenc* Predict* Factor* Association* Risk* Relation* Correlat* Aggregat*
CINAHL Headings:	CINAHL Headings:	CINAHL Headings:	CINAHL Headings:
Parents (Exp)	Pain (Not Exp) Chronic Pain (Exp) Fibromyalgia (Not Exp) Irritable Bowel Syndrome (Not Exp) Arthritis (Not Exp) Osteoarthritis (Not Exp) Headache (Not Exp) Migraine (Not Exp) Neuralgia (Not Exp) Peripheral Nervous System Diseases (Not Exp) Complex Regional Pain Syndromes (Not Exp)	Child (Exp) Child, Preschool (Not Exp) Adolescence (Exp) Infant (Exp) Young adult (Not Exp)	Predictive Research (Not Exp) Correlational Studies (Not Exp) Risk Assessment (Not Exp)

Table A3. Search Terms: EMBASE

Parental Terms	Pain Terms	Child Terms	Relationship Terms
Title/abstract:	Title/abstract:	Title/abstract:	Title/abstract:
Parent* Mother* Maternal Father* Paternal Family Families Caregiver* Care-giver Care-givers	Pain* Fibromyalgia Irritable bowel syndrome Arthrit* Osteoarthrit* Headache* Migraine* Neuralgi* Neuropath* Complex regional pain syndrome	Child* Adolescen* Teen* Youth* Infant* Young adult Young adults	Impact* Influenc* Predict* Factor* Association* Risk* Relation* Correlat* Aggregat*
Emtree Terms:	Emtree Terms:	Emtree Terms:	Emtree Terms:
Parent (Exp)	Pain (Not Exp) Chronic Pain (Not Exp) Fibromyalgia (Not Exp) Irritable Colon (Not Exp) Arthritis (Not Exp) Osteoarthritis (Not Exp) Headache (Not Exp) Migraine (Not Exp) Neuralgia (Not Exp) Peripheral Neuropathy (Not Exp) Complex Regional Pain Syndrome (Not Exp)	Child (Exp) Preschool Child (Not Exp) Adolescence (Exp) Infant (Exp) Young adult (Not Exp)	Predictive Research (Not Exp) Correlational Studies (Not Exp) Risk Assessment (Not Exp)

Table A4. Search Terms: PsycINFO

Parental Terms	Pain Terms	Child Terms	Relationship Terms
Title/abstract:	Title/abstract:	Title/abstract:	Title/abstract:
Parent* Mother* Maternal Father* Paternal Family Families Caregiver* Care-giver*	Pain* Fibromyalgia Irritable bowel syndrome Arthrit* Osteoarthrit* Headache* Migraine* Neuralgi* Neuropath* Complex regional pain syndrome	Child* Adolescen* Teen* Youth* Infant* Young adult*	Impact* Influenc* Predict* Factor* Association* Risk* Relation* Correlat* Aggregat*
Subject Terms:	Subject Terms:	Subject Terms:	Subject Terms:
Parents (Exp)	Pain (Not Exp) Chronic Pain (Not Exp) Fibromyalgia (Not Exp) Irritable Bowel Syndrome (Not Exp) Arthritis (Not Exp) Headache (Not Exp) Migraine Headache (Not Exp) Neuralgia (Not Exp) Peripheral Neuropathy (Not Exp) Complex Regional Pain Syndrome (Type I) (Not Exp)	Offspring (Exp)	Risk Assessment (Not Exp) Risk Factors (Not Exp) Prediction (Not Exp) Statistical Correlation (Not Exp)

Supplementary Materials - Appendix B

Summary of Meta-Ethnography Results **for the Three Included Qualitative Studies**

	Evans & de Souza, 2008	Umberger et al., 2013	Umberger et al., 2014
Sample	<ul style="list-style-type: none"> • 21 children and their 16 mothers with chronic pain • 7 girls, 14 boys, ages 6-12 ($M = 8$ years, 9 months) 	<ul style="list-style-type: none"> • 30 young adults ages who identified as having lived during their adolescence with a parent suffering with chronic non-cancer pain (CNCP) • 21 females, 9 males, ages 18-21 	
Data Collection	<ul style="list-style-type: none"> • Interviews conducted in participants' homes; mothers and children interviewed separately • "Themes explored in interviews with children included: mother's health and pain, how children knew about their mother's health, children's feelings about their mother's pain, and the child's health." (p. 492) 	<ul style="list-style-type: none"> • Interviews conducted at university or other accessible public places • "Participants were interviewed once and encouraged to tell their stories about what it was like growing up with a parent who suffered with CNCP and their day-to-day management strategies." (pg. 346, Umberger et al., 2013) 	
Setting	<ul style="list-style-type: none"> • Small city in New Zealand 	<ul style="list-style-type: none"> • Ohio, USA 	
Types of parental pain	<ul style="list-style-type: none"> • Lupus ($n = 1$), arthritis ($n = 1$), polycystic ovary syndrome ($n = 1$), migraine ($n = 2$), repetitive strain injury ($n = 2$), spinal pain ($n = 9$) 	<ul style="list-style-type: none"> • 18 had mothers with CNCP, 12 had fathers with CNCP • Arthritis ($n = 12$), fibromyalgia ($n = 6$), migraine headache ($n = 5$), herniated disc ($n = 4$), degenerative joint disease ($n = 3$), neuropathy ($n = 3$), fractured vertebrae ($n = 3$), carpal tunnel ($n = 2$), Crohn's disease ($n = 2$), chronic regional pain syndrome ($n = 1$), multiple sclerosis ($n = 1$), paraneoplastic syndrome ($n = 1$) 	
Study perspective	<ul style="list-style-type: none"> • Developmental gains-and-loss perspective • Strengths perspective 	<ul style="list-style-type: none"> • Grounded theory 	
Research questions	"a) understand the challenges that mothers with chronic pain face, as	"a) how do adolescents manage living with a parent suffering with CNCP? And b) what environmental factors help or hinder this	

	well as their children; b) examine how mothers and children overcome these challenges; and c) discuss maternal chronic pain as a two-way reciprocal process, with mothers and children impacting on the well-being of each other.” (pg. 491)	process?” (pg. 345, Umberger et al., 2013)	
Data analysis	<ul style="list-style-type: none"> • Qualitative conceptual framework approach • Categories determined before the interviews, but also included topics participants discussed outside of those categories 	<ul style="list-style-type: none"> • Grounded theory • Constant comparative methods • 3 levels of coding, 6 investigators involved in coding 	
Themes			
Developing independence	Children developed increased independence and cared for themselves and their mothers.	Some adolescents described having to endure hardships related to developing independence (e.g., having to fulfill a parental role before being ready to do so). Some also used their independence as means of coping with their parent’s pain.	Participants described “life lessons”, including “growing up too fast is not such a good thing”. They described the hardships of having to be independent from an early age.
Developing compassion or empathy for others	Mothers and children reported that children were very caring and helped with household tasks. Most children reported enjoying being able to help their mother.	Some adolescents were able to empathize with their parents, including trying to understand the motives behind their parents’ behaviours and the complexity of their parents’ pain problems.	Participants described learning the “life lesson” that “it is important to look beneath the surface”. They became able to empathize with others, especially those experiencing adversity.
Learning about health and coping	Children learned more about health, illness, their own bodies, and how to handle stress because	Not described in this analysis	“Many participants described themselves as being “better off” because of having learned some

	of having a mother with chronic pain.		sort of “life lesson” while growing up with a parent with chronic pain.” (pg. 6)
Missing out	Activities were often missed because of the mother’s chronic pain. Some children expressed anger because of the experiences they had to miss out on because of their mother’s chronic pain.	Some participants described experiencing strong negative emotions related to the loss of their childhood and authentic parents.	Participants learned the life lesson that “sometimes you have to accept that ‘it’s just the way it is’” and came to accept missing out on certain things.
Effects on emotional health	Children became “clingy” and experienced anxiety and sadness related to their mother’s pain. In several cases, the extent of the child’s anxiety regarding their mother’s pain was not known to their mother.	Participants experienced deep, negative emotions regarding their experiences with parental chronic pain. Others worried about whether their parent would be present for important events in the future.	Some participants experienced anxiety and worry related to not knowing whether their parent’s pain problems were life-threatening. Others worried whether they would experience chronic pain themselves.
Struggles with communication	As described above, in some cases mothers were unaware of the worries their children had regarding their mother’s pain.	Some adolescents distanced themselves emotionally and physically from their parents, not sharing their feelings about their parent’s pain. Some were able to seek emotional support from other family members or friends, while others were not.	Few participants described receiving clear information about their parent’s pain. Many avoided initiating conversations about pain because they believed that it would cause their parent more pain and distress. They described trying to construct meaning about their parent’s pain on their own.