A Developmental Perspective of Influences on the Onset and Early Trajectory of Chronic Pain in Children Attending Physiotherapy in Primary Health Care Settings: An Integrative Review

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ABSTRACT

Chronic pain in childhood leads to long-term social, emotional, physical, and financial impacts, with lifelong heightened pain sensitivities. The prevalence of chronic pain in children has increased over the past 20 years. Physiotherapy is often the place of initial intervention for a child in pain, and initial treatment is predictive of chronic pain and can predict outcomes. Children's pain experiences and responses are thought to be more modifiable than those of adults; therefore, quality physiotherapy interventions have the potential to positively influence the onset and early trajectory of chronic pain. Using a childhood developmental lens, this integrative review aimed to draw together research literature about biopsychosocial factors influencing chronic pain onset and early trajectory in primary-school-aged children. The results of the review contribute novel understandings for physiotherapists in primary healthcare. A range of biopsychosocial concerns including age at onset and pain duration, neuro-biological aspects, psychological influences, parental impacts, sleep quality, trauma history, and stress interact with children's learning and development. Taken together, these factors have potential to influence chronic pain onset and early trajectory in children. Adopting a holistic understanding of childhood development in the early management of childhood chronic pain would promote positive physiotherapy interventions and may avoid pathologising what is developmentally normal.

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INTRODUCTION

Childhood is a period of extensive growth and learning. When physiotherapists are working with children and young people there needs to be an understanding of the developmental physical, cognitive, and psychosocial changes that children undergo (Cech et al., 2019). Despite this need, current literature informing practice within children with chronic pain gives little consideration to developmental factors that can influence the onset and trajectory of this condition. Additionally, research to date has typically focused on various, discrete aspects of children's pain including sleep quality (Kanstrup et al., 2014; Pavlova et al., 2020), gender influences (Kaczynski et al., 2009) and threat or fear responses (Flack et al., 2017; Heathcote et al., 2017). A more cohesive picture of the influences is needed to support practice.

Studies suggest a trend towards increasing prevalence of chronic pain in childhood (Coffelt et al., 2013; King et al., 2011; Roy et al., 2022). The term chronic pain typically describes pain of longer than 3 months duration or pain that has exceeded expected tissue healing timeframes, without evidence of ongoing injury or biological markers (Abu-Arafeh & Abu-Arafeh, 2016; Campos et al., 2011; Collins et al., 2017). Chronic pain is not just continuous pain, and children may also experience intermittent, yet reoccurring episodes of pain over long periods

(Bhatia et al., 2008; King et al., 2011). Chronic pain is often, but not always, precipitated by an injury or tissue damage. However, the onset and progression of a chronic pain course is complex, and a wide range of biopsychosocial factors may contribute to its presentation (Swain & Johnson, 2014). A consistent pattern of childhood chronic pain negatively affects pain sensitivities and nociceptive thresholds into adulthood, creating a lifelong pain influence (Bhatt et al., 2020; Hassett et al., 2013; McClain & Suresh, 2009; Tan et al., 2009).

The biopsychosocial model is widely accepted to guide clinical practice in people with chronic pain (McGrath et al., 2014). The model provides a theoretical framework whereby the complex, non-linear interplay of an individual's mind, body, and social environment are identified as valid contributors to a person's overall health experience (Engel, 1977). The separation of chronic pain into compartments of physical or psychological factors has now been systematically excluded from research and clinical practice guidelines (Bursch et al., 1998). Nonetheless, a dualistic concept of mind–body remains, and seeking an organic and physical explanation for a child's pain has been thoroughly embedded within healthcare (Dell'Api et al., 2007; Hinton & Kirk, 2016). Unfortunately, healthcare practitioners' disregard for a more holistic view of pain management contributes to tension in the practitioner–family relationship (Hinton & Kirk, 2016).

Frustrations are reported by children, parents, and their healthcare providers when endeavouring to understand and manage chronic pain conditions (Hinton & Kirk, 2016). Dismissal of children's pain concerns is extensively reported in qualitative research (Carter, 2002; Dell'Api et al., 2007; Newton et al., 2013; Quintner et al., 2008). A lack of training in managing child-specific pain conditions (Defenderfer et al., 2018), a lack of knowledge of developmental consequences of pain (Bhatia et al., 2008), and a lack of clinical guidelines are highlighted as reasons for delays in diagnosis and subsequent tertiary referrals for children with chronic pain (Abu-Arafeh & Abu-Arafeh, 2016; Hinton & Kirk, 2016). Prior to arriving at tertiary, interdisciplinary team management for a child's chronic pain, families have reported low expectations for future interventions due to disappointment from earlier healthcare interactions (McGrath et al., 2014).

Primary care (PC) practitioners, such as physiotherapists, are key to managing the onset and interrupting the trajectory of chronic pain for this group. The effectiveness of the first treatment is predictive of reducing chronic pain risk in children (Simons et al., 2018; Wager et al., 2019). Additionally, children's pain perception is thought to be relatively more "plastic" than adult populations, indicating that in the preliminary stages their pain may be more modifiable with appropriate intervention (Bhatt et al., 2020; Campos et al., 2011; Zernikow et al., 2018). Furthermore, a longer time to diagnosis has been associated with a greater level of functional disability (Carter, 1998; Tian et al., 2018). Simply, untreated or poorly managed pain in children is a risk factor for chronicity (Finley et al., 2014).

Developmental theory provides frameworks for explaining a process of change in people over time (Miller, 2016). These theoretical frameworks are relevant for understanding children's responses to pain and how the responses are shaped over time. Human development involves not only biological processes, but also environmental experiences and social reinforcement (Bergen, 2008). Tenets from social cognitive theory (SCT) (Bandura, 1986), sociocultural developmental theory (SCDT) (Vygotsky et al., 1978), and understandings about children's neuropsychological development may better our understanding of ways that children learn about pain experiences and their responses to factors that potentially influence pain.

Social cognitive theory

Social cognitive theory (SCT) highlights that children learn, not solely through teaching, but by observing and modelling others' behaviour (Bandura, 1986). SCT focuses on interactions between the situation, emotions, social interactions, and people's behaviour and how these things are perceived by the child (Miller, 2016; Smith, 2013). Learnings are appraised over time, and with reinforcement, particular behaviours and responses develop (Miller, 2016).

Self-efficacy is a central acquisition of social cognitive observations (Miller, 2016), enabling a child to believe in their abilities and determine their capacity to cope with adversities (Page & Blanchette, 2009) or threat (Steck & Steck, 2016), such as pain. Self-efficacy and resilience only develop as positive personality characteristics if the child can learn the skills required through social learning constructs (Bandura, 2003; Miller, 2016; Steck & Steck, 2016).

SCT is a key framework to guide understanding of the influences on a child's pain within their family and wider social system (Levy et al., 2007). Such influences are often hypothesised as the reason for the clustering of chronic pain conditions within a family (Stone et al., 2018).

Sociocultural developmental theory

Sociocultural developmental theory (SCDT) varies from SCT in acknowledging the central role of adults and cultural contexts in children's development. SCT holds that a child develops skills through interactions and problem solving with more cognitively advanced individuals around them (Koenig & Sabbagh, 2013; Miller, 2016; Vygotsky et al., 1978). The "culture context" relates to the wider values, beliefs, and history associated with the child's environment and their social setting (Miller, 2016; Vygotsky et al., 1978). In this way, children's' understandings about the diverse facets of a pain experience may be seen as being actively shaped by their sociocultural environment, including their ideas about the causes of pain, adoption of management strategies, their responses, and communication.

Neurobiological and neuropsychological development

Neurobiological and neuropsychological developmental changes are also an important basis for a child's learning and development, and likely play a role in the ways they respond to pain over childhood. Children's brain regions have been found to have lower levels of connectivity that improve throughout development, and cortical changes associated with neuroplasticity result in, and support, adaptive learning, and neural network modifications (Fine & Sung, 2014). Exaggerated responses to pain have been reported due to immaturity of neurotransmitters, increased neural excitability, and less neural inhibitory pain control (Hathway, 2014). From a developmental neuropsychology perspective, these exaggerated responses may, therefore, be a normal response due to still-developing executive brain functions needed to exhibit stability over supraspinal controls (Feinstein et al., 2017; Hathway et al., 2012). Likewise, children's cognitive and emotional understanding of pain, their appreciation of others' pain behaviour, and insights into the social implications of pain behaviours are not refined until early adolescence (Esteve & Marguina-Aponte, 2012).

Understandings from SCT, SCDT, neurobiological, and neuropsychological theories collectively suggest that various developmental influences will be relevant to the onset and early trajectory of chronic pain in children. This review aimed to apply a childhood developmental lens to draw together and critically analyse the existing research literature about the biopsychosocial factors potentially influencing chronic pain onset and early trajectory in children. An integrative review methodology (Russell, 2005; Whittemore & Knafl, 2005) was employed to synthesise and gain new insights into the literature through application of developmental theoretical understandings.

METHODS

A thorough search strategy was implemented, utilising online databases CINAHL, MEDLINE, Scopus, and AMED. Published studies from January 2000 to January 2021 were included. A manual search of the reference lists of the included studies was also undertaken. The search terms Boolean operators, limits, and numbers of hits are presented in Table 1.

Inclusion and exclusion criteria

Using the Covidence tool (Veritas Health Innovation, 2020), all identified published studies were initially screened by their title and abstract (AM & MJ), then a full text review was conducted to check relevance against the inclusion and exclusion criteria outlined in Table 2.

In line with the need to draw together relevant but often disparate information, the inclusion criteria were broad to generate new insights about the topic from diverse perspectives (Whittemore & Knafl, 2005). Not only were studies included that specifically explored relationships between biopsychosocial factors and pain outcomes, but also those that shed light on the prevalence and characteristics of factors that could potentially influence pain.

Search outcomes

The initial search provided 678 abstracts and 47 articles were included in the final review.

Data evaluation and analysis

The focus of an integrative review is on merging diverse understandings about a topic to provide a more cohesive knowledgebase (Torraco, 2016). Systematic critical appraisal is not always the focus of an integrative review (Kirkevold, 1997; Whittemore & Knafl, 2005). As the present review aimed to use a child development theoretical lens to draw together and critically analyse research literature about the factors influencing chronic pain onset and trajectory in children, all included studies were analysed and reported (Torraco, 2016; Whittemore & Knafl, 2005). While a formal, critical appraisal or scoring system was not applied, the methodological quality of the diverse

research was critically considered in light of design features and ability to represent the population (Whittemore & Knafl, 2005), and is discussed in this report.

Information was extracted from the articles about the study designs, population demographics, pain duration, and findings relevant to answering the research question (Table 3). Similar themes in the findings were grouped together in consideration of their focus and were then critically considered and integrated with paediatric developmental literature (Appendix A).

REVIEW FINDINGS

Studies that met the inclusion criteria were primarily observational, with 20 cross-sectional, 13 cohort, seven case control, one epidemiological prevalence study, and three individual case reports included for review. Three qualitative studies were also obtained (Carter, 2002; Dell'Api et al., 2007; Pate et al., 2019) and provided the voice of chronic pain experience from children themselves and their parents.

There were various limitations shared by the majority of studies in this review. Limitations included use of parent proxy-reports, a risk of recall bias, and cross-sectional methodologies that limited insights into changes in children's pain trajectories over time. Many studies did not provide or analyse data about variables such as age at onset of chronic pain, pain duration (Lynch et al., 2007), and interventions children may have received. Generalisability of study findings to primary health care settings and to younger children was constrained; most studies were undertaken in tertiary care and had a low representation of children under the age of 10 years, with some studies containing primarily adolescent data (Table 3).

Table 1 *Literature Search Strategy*

Search terms	Limits used	Database and sites searched	Number of hits
Child* OR a pediatric* OR paediatric* OR adolescen*	English language	EBSCO-	
OR youth OR "school age*" OR "school-age" OR		Cinahl	
kid* OR young		Medline	550
AND		Scopus	119
Risk* OR predict* OR influence* OR contribut* OR factor* OR cause*		AMED	3
AND			
"persistent pain" OR "chronic pain" OR "complex regional pain syndrome" OR "neuropathic pain" OR "pain syndrome*" OR "secondary hyperalgesia" OR "central hyperexcitability" OR CRPS OR "somatoform pain" OR "unexplained pain"			
NOT			
Scoliosis OR "juvenile idiopathic arthritis" OR "cerebral palsy" OR haemophilia OR hemophilia OR "sickle-cell" OR cancer			
		Manual search	3

^a AND, NOT, and OR are the Boolean operators that were used in the search.

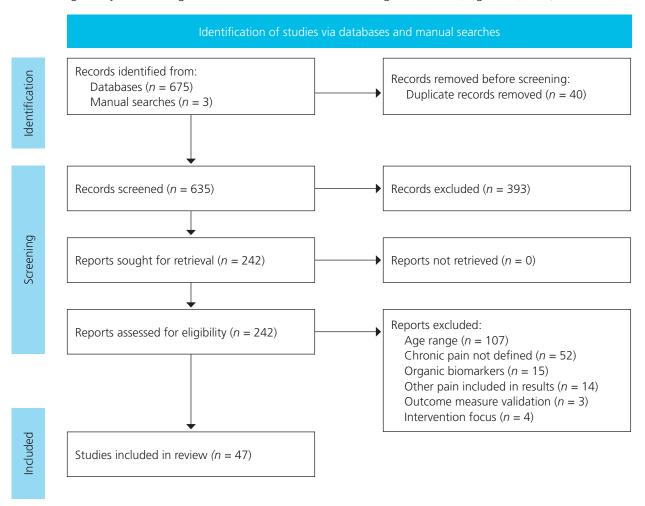
^{*}Denotes truncation.

Table 2 *Inclusion and Exclusion Criteria*

Inclusion criteria	Exclusion criteria
Primary research	Secondary studies
Chronic or recurrent pain > 3 months or met strict diagnostic	Organic, biomarkers for pain
criteria ^a	Scoliosis surgery, juvenile idiopathic arthritis, cerebral palsy,
Unknown organic aetiology	haemophilia, and sickle-cell disease.
Participants aged 5–11 years	Studies involving adults (>18 years of age)
Parents with children aged 5–11 years	Intervention only studies

^a Diagnostic standards included: Complex regional pain syndrome (CRPS), International Association for the Study of Pain criteria (Stanton-Hicks et al., 1995) or Budapest Criteria (Harden et al., 2007); Juvenile primary fibromyalgia syndrome according to the Yunus and Masi criteria (Yunus & Masi, 1985); and Functional abdominal pain (FAP) as per the International Headache Society classification (ICHD) (Olesen, 2018) and Rome III criteria for FAP (Shih & Kwan, 2007).

Figure 1 *PRISMA Diagram. Systematic Diagram of Identification of Studies for Integrative Review (Page et al., 2021).*



Influences on the onset and early trajectory of children's chronic pain

Analysis of the study findings identified a number of potential child-related and social influences on the onset and early trajectory of children's chronic pain. The influences either reflected or had implications for children's development (Figure 2).

Child age-related influences

Data about children's ages can provide a general indication of their development (Bergen, 2008), but age was only analysed as a factor in a small number of studies, with mixed findings reflecting different study foci. In one study, children's age showed little relationship to the extent of functional disability as measured through physical domains of a quality of life

Table 3 Summary of Included Articles

Author (date)	Methodology Participant	Participants N (sex)	Pain duration	Developmental framework	Primary findings
	recruitment Country	Age Diagnosis			
Abu-Arafeh and Abu- Arafeh (2016)	Prospective observational study Community sample through Scottish paediatric surveillance Scotland	26 5.5–15.4 (M = 11.9) years CRPS	Not stated	<u>0</u>	Average 22 missed school days, 23 stopped participating in sports, 19 cancelled social engagements. CRPS incidence measured 1.2/100,000. Legs more frequently affected than arms with second body site present in 40%. Mean onset 11.9 years, primary onset after trauma, and affecting girls 3:1 ratio. Right side more affected than left. Higher functional disability and somatic symptoms. Normal depression and anxiety scores. Parental effects of work adjustments and cancelling holidays.
Agrawal et al. (2009)	Cohort retrospective chart review Outpatient tertiary pain clinic USA	32 11–15 years CRPS	Not stated	<u>0</u>	Movement disorders/dystonia present with CRPS at similar rates to with adults. Mainly lower limb affected.
Akbarzadeh et al. (2018)	Cross-sectional, convenience sampling Two outpatient tertiary clinics and a private clinic	132 children (72 girls, 60 boys) 212 parents (120 mothers, 92 fathers) <i>M</i> (<i>SD</i>) = 9.83 (2.77) Chronic headaches	20.56 months	<u>0</u>	Differences between maternal and paternal pain catastrophising with mothers higher reporting for their child on pain intensity, anxiety, depression in relation to their child's headache pain. Greater parental catastrophising mothers than fathers, increasing attention to their child's pain. Parents central mediators to their child's pain.
Bayle-Iniguez et al. (2015)	Case-control, retrospective Single centre outpatient clinic Not stated	73 (64 girls, 9 boys) 5–16 (<i>M</i> = 11.5) years CRPS	Not stated Time to diagnosis, 0–120 months (M = 14.2)	O _N	Excellent school results ($OR = 8.4$, 95%CI), perfectionist behaviours ($OR = 4.3$, 99.5% CI); Issues falling asleep ($OR = 6.9$, 95% CI); Anxiety and CRPS strongest association ($OR = 44.9$, 95% CI). Psychosocial stress in 51% cases including family conflict, medical issue or death of family member, school problems, sexual abuse. Physical trauma 49% (92% minor injuries). CRPS type 1 mostly affecting younger pre-adolescent age group, lower limb (89%) and predominantly girls.
Beeckman et al. (2020)	Cohort study Two outpatient tertiary clinics USA	65 (54 girls, 11 boys) M (SD) = 14.41 (1.95) years Mixed chronic pain conditions	M (range) = 27.05 (3–96) months	ON	Pain related fear predicts function better than pain intensity. Possible bidirectional relationship between pain related fear and catastrophising. Psychological flexibility predicted lesser pain levels and less avoidance behaviours.
Birnie et al. (2020)	Cross-sectional Outpatient pain clinic USA	108 children 8–12 years 340 adolescents 13–18 years <i>M</i> (<i>SD</i>) = 14.57 (2.38) years Mixed chronic pain conditions	Children, $M =$ 2.53 years Adolescents, $M(SD) = 30.41$ (35.04) months	Analysis of children and adolescents separately	Links created of integrative model of parental health with fear-avoidance in children with chronic pain. Greater pain interference associated with increased age. Poorer parental global health indirectly related to child pain interference through higher parent pain catastrophising, then greater parent protective behaviours and child pain catastrophising.

Author (date)	Methodology Participant recruitment Country	Participants N (sex) Age Diagnosis	Pain duration	Developmental framework	Primary findings
Carter (2002)	Qualitative research Outpatient clinic Nominated families from nurse practitioner Not stated	3 7–13 years M = 12.7 years Mixed chronic pain conditions	≥ 3 months	O Z	Main themes: (a) searching for a diagnosis, (b) professional dismissal, and (c) medical communication. Families less focus on child's pain but more on difficulties with healthcare professionals. Parents used to communicate about their child's pain than the child themselves.
Conte et al. (2003)	Case control study Single outpatient clinic USA	16 with JPFMS 16 with arthritis 16 healthy controls 7.4–17.4 years	3–36 (<i>M</i> = 12) months	O _N	JPFMS group lower mood, high anxiety and depression, irregular daily structure, withdrawal, somatic complaints, highly distractible when compared to the other two group. Significantly greater behavioural issues. Parents of children with JPFMS had higher levels of depression, anxiety. Parents reported for their children greater pain sensitivity than the other two groups.
de Tommaso et al. (2017)	Cross sectional study Single outpatient tertiary clinic Italy	151 8–15 years Chronic headache or migraine	≥ 3 months	O Z	Muscle pain/peri-cranial tenderness intensity related to reduced duration of sleep. Loss of sleep may contribute to central sensitisation. Allodynia symptoms correlated to anxiety.
Dell'Api et al. (2007)	Qualitative semi- structured interviews Single outpatient tertiary clinic Canada	5 (2 boys, 3 girls) 10–17 years Mixed chronic pain	6 months to ≥ 4 years	0 N	Main theme: Participation limitations. Main subthemes: Seeking understanding of pain, invisibility of pain, impact of encounters, future fears, negative perceptions of pain; Children felt disbelieved and dismissed. Lack of information regarding their pain, more likely to perceive pain as life threatening in children 10–12 years.
Dunn et al. (2011)	Cohort study Prospective, community sample USA	1336 11 years–11 years 10 months, initial sampling Prevalence and trajectories of chronic pain measured	Not stated, observational prevalence study	Pubertal development scale and age controled	12% of sample had persistent pain for at least one pain site. Persistent pain group primarily female, high baseline levels somatisation and depression at age 11 and at end of study at 14 years.
Erpelding et al. (2014)	Case control Outpatient clinic USA	12 (9 females, 3 males) 10–17 years M = 14.1 years with unilateral CRPS Healthy controls (9 females 3 males) M = 13.8 years	M = 13 months	O Z	Habenula involvement in modulating responses of stress, reward-punishment and pain modulatory process, which may contribute to CRPS symptoms including affective, cognitive, pain inhibition and motor processing. Reductions in functional magnetic resonance imaging between Hb and pain inhibitory systems.
Fales et al. (2014)	Cross sectional study Outpatient clinic USA and Canada	210 (73.9% female) 10–17 years M (<i>SD</i>) = 14.23 (1.59) Mixed chronic pain conditions	≥ 3 months	O Z	Low agreement between child and parent in regard to miscarried helping. Child and parent more likely to report miscarried helping with dysfunctional and less cohesive family network reports, greater reports of child depression.

Author (date)	Methodology Participant recruitment Country	Participants N (sex) Age Diagnosis	Pain duration	Developmental framework	Primary findings
Finniss et al. (2006)	Case report Primary care Australia	Single case study 10-year-old male CRPS	7 weeks	ON ON	Pain, colour, swelling and temperature changes in foot. Nil trauma.
Flack et al. (2017)	Cross sectional study Inpatient and outpatient Germany	40 11–18 years Chronic headache and chronic abdominal pain	IHSC, ICD-10 and Rome III criteria	ON	Proximal interoceptive sensations caused greater fear-avoidance in chronic abdominal pain group. Lower task performance time in the associated threat task in both groups. Fear-avoidance behaviours were elicited when benign-threat tasks are applied near to the primary pain region. Attention bias is anticipatory, not responsive.
Heathcote et al. (2017)	Case control study Outpatient tertiary pain clinic, patient group Two secondary schools, controls UK	66 (55 female, 11 male) 10–18 years M (5D) = 13.97 (2.13) years 74 controls (42 female, 32 male) 11–18 years M (5D) = 14.95 (1.71) years Mixed chronic pain conditions	5-170 (M = 45.7) months	O _N	Interpretation bias of ambiguous body threat tasks in chronic pain group, more likely to perceive threat. 87.9% had widespread musculoskeletal pain vs 12.1% single location.
Ho et al. (2009)	Cross-sectional study Consecutive referrals tertiary pain service Canada	57 (46 females, 11 males) 8–18 years M (5D) = 14.64 (2.39) years Mixed chronic pain conditions	M (5D) = 46.98 (34.92) months	ON	Above average scores on all cognitive and academic assessment measures.
Huguet and Miró (2008)	Prevalence cross- sectional study Community sample Spain	561 8–16 years M (SD) =11.89 (2.00) years	Prevalence study	<u>0</u>	Lower social QoL scores; Lower limb chronic pain more common in boys (57% vs 19.65%) and multiple site chronic pain more common in girls (50.6% vs 22.2%). Lower physical QoL scores. Abdominal pain, headache, and lower limb complaints most common location of chronic pain. Prevalence of chronic pain increased with increasing age: Chronic pain, M (SD) = 11.49 (2.08) years; Without chronic pain, M (SD) = 10.53 (1.86). Age did not impact functional disability or QoL measures in chronic pain sample. 37.3% with chronic pain, 29.3% had pain in multiple sites. Only 5.1% with moderate/severe problems.
Hunfeld et al. (2002)	Cross sectional study Community sample from larger prevalence study Netherlands	85 (56 girls, 29 boys) 5–11 years <i>M</i> (<i>SD</i>) = 8.4 (2.1) years (boys) <i>M</i> (<i>SD</i>) = 8.4 (2.4) years (girls) Unexplained chronic pain	M (5D) = 3.1 (2.3) years	O N	School absence, days per month, $M = 0.5$ day, no gender differences. Abdominal, limb pain and headaches most common reported areas of pain. High intensity of perceived pain of the child had a higher social impact on family, especially mothers. Chronic pain impact on families reported to be mild.

Author (date)	Methodology Participant recruitment Country	Participants N (sex) Age Diagnosis	Pain duration	Developmental framework	Primary findings
Kaczynski et al. (2009)	Cross sectional study Outpatient clinic USA	266 (female 66.5%) 8–17 years M (SD) = 13.3 (2.55) years Chronic abdominal pain and headaches	M (SD) = 32.68 (32.53) months	ON	Pain rating associated with depression in boys and girls; related with depression and anxiety in girls, not boys. Depression more readily reported by girls than boys. Passive coping linked to increased internalising behaviours. Protective parenting correlated with functional disability in boys not girls, but not statistically significant. Internalising symptoms associated with protective parenting.
Kanstrup et al. (2014)	Cohort study Outpatient pain clinic Sweden	154 10–18 years M (SD) = 14.57 (2.02) years Mixed chronic pain conditions	3–192 months <i>M</i> (<i>SD</i>) = 52.46 (43.44) months	Age correlations only	31.5% reported one per week absence from school due to pain, and 44.3% reported wide or total absence from school. Pain is a predictor of functional disability with insomnia a mediator. Insomnia contributed more variance than pain intensity to the association of depression and functional disability. Girls reported higher prevalence of depression ($M = 25.6 \text{ vs } 20.0; p = 0.011$) and depression symptoms increased with age ($p = 0.038$). Pain duration longer in boys than girls ($M = 67.0 \text{ vs } 47.7; p = 0.022$). No significant age-related differences between functional disability, intensity or duration of pain.
Kashikar- Zuck et al. (2008)	Cross sectional study Outpatient pain clinic USA	76 (female 86.8%) 11–18 years <i>M</i> = 14.89 years JPFMS	≥ 3 months	ON	67.1% had a current psychiatric disorder and 30.1% had more than one disorder. 71.2% had a history of psychiatric disorder. Most common diagnosis of anxiety.
Kashikar- Zuck et al. (2010)	Cross sectional study Outpatient pain clinic USA	102 (87.3% female) 11–18 years <i>M</i> (<i>SD</i>) = 14.96 (1.82) years JPFMS	M (SD) = 35.77 (27.83) months	No	School days missed, $M = 2.88$ days/month; 12.7% home- schooled due to their JPFS. High level anxiety conditions. 24% attentional disorder, 19% depression. High level of school absence linked to greater depression ($p = 0.03$). Mothers reported $M = 5$ pain conditions.
Konijnenberg et al. (2005)	Cross sectional study Outpatient pain clinic Netherlands	149 $8-18$ years $M(SD) = 11.8(2.6)$ Unexplained chronic pain	M (range) = 12 (3–144) months	No	Highest social functioning inference with headache (50.9 headache, 73.48 abdominal pain, 77.7 musculoskeletal pain). Children who had a poor view of their own health had significantly higher levels of pain related impairment and this was predictive of significant impairment.
Lebel et al. (2008)	Cohort study Outpatient tertiary pain clinic USA	8 (all female) 9–18 years M (SD) = 13.5 (1.6) years CRPS, lower extremity unilateral	M (SD) = 13.3 (2.4) months	O _N	Increased BOLD signal in parietal lobe, bilateral primary sensory cortex, anterior and middle cingulate, anterior insula. Decreased BOLD signal in frontal lobe and parietal lobe, middle cingulate, middle temporal lobe, parahippocampus and hippocampus. Decreased signals in pain inhibition pathways. Contralateral/unaffected limb had almost double the increased BOLD signal and few regions showing decreased BOLD signal. Findings were similar when pain state had resolved. Notable central processing changes that are maintained after symptom resolution may be related to the developmental plasticity and have long lasting effects.

Author (date)	Methodology Participant recruitment Country	Participants N (sex) Age Diagnosis	Pain duration	Developmental framework	Primary findings
Libby and Glenwick (2010)	Cross sectional study Outpatient tertiary rheumatology clinics USA	57 10–18 years <i>M</i> = 15.5 years JPFMS	Not stated	ON ON	School absence 2.5 days in past month (range 0–31 days). Daily hassles not related to pain ratings. Poor pain ratings, QoL, and depression scores with catastrophising. Self-efficacy positively influenced QoL, catastrophising, pain, and depression scores. Social support from family and peers improved QoL and improved pain. 16% had a parent with fibromyalgia, 56% had a parent who had chronic pain. No significant differences existed between age group analysis of 10–14-year-olds and 15–18-year-olds and variables of pain, perceived social support, coping strategies, depression, or self-efficacy.
Linnman et al. (2013)	Cohort study Outpatient pain clinic USA	8 9–18 years <i>M (SD)</i> =13.5 (1.6) years CRPS	Not stated	ON	Symptomatic and persistent cortical reorganisations – significant increased connectivity alterations anterior cingulate, postcentral gyrus, putamen, amygdala, caudate, and thalamus. Alterations in multiple sensory, emotional and cognitive processing regions. The amygdala showed transient increased connectivity in BOLD signal analysis to the primary cingulate cortex.
Logan et al. (2013)	Retrospective, cross- sectional case control study Single outpatient pain clinic USA	101 7–18 years M (SD) = 13.41 (0.24) years CRPS Comparisons 103 abdominal pain, 291 headache, 119 back pain	CRPS, 13 months Headache, 32.60 months Abdominal pain, 28.14 months Back pain, 23.25 months	ON	CRPS group fewer missed school days than other groups. CRPS group no greater anxiety, depression and relative to normative values. More females with CRPS proportionately than the other pain groups. CRPS group reported more somatic symptoms.
Lynch et al. (2007)	Cross sectional study Outpatient pain clinic USA	272 (66% girls) 8–18 years Mixed chronic pain	3–24 months	Analysis of children and adolescents separately	Boys more likely to participate in physical activity as a distraction from pain. Girls sought social support more than boys and expressed their pain and feelings more. Older children had longer pain duration and become increasingly distressed and anxious with the long duration of their pain. Younger children rely heavily on social supports for pain related strategies and affirmations.
Noel, Beals- Erickson et al. (2016)	Cross sectional study Outpatient pain clinic USA	46 caregivers of children (41 mothers, 1 grandmother) aged 10–18 years M (5D) = 13.51 (2.00) years Mixed chronic pain	3 to ≥ 60 months	O _N	Orientation with acceptance of diagnosis unresolved diagnosis beliefs showed correlation to resilience or distress cluster groups of parents. Parents in resilience narrative cluster were more likely to show humour, positive outlook, and benefits with their child's pain. Parents reported higher levels of depression and anxiety. Extent of the child's functional disability did not explain the degree of parental distress levels.

Author (date)	Methodology Participant recruitment Country	Participants N (sex) Age Diagnosis	Pain duration	Developmental framework	Primary findings
Noel, Wilson et al. (2016)	Case control study Outpatient pain clinic USA	95 10–17 years M (SD) = 15 (2.1) years Chronic pain 100 controls M (SD) = 14.3 (2.1) Mixed chronic pain conditions	≥ 3 months	O _N	QoL lower in children with chronic pain compared to controls. Chronic pain group reported death and abuse as their most traumatic life event.
Pas et al. (2019)	Case control study Inpatients Belgium	39 FAP, 36 controls 6–12 years <i>M</i> = 9 years	M = 24 months $Mdn = 28$ months	O _Z	Pressure pain thresholds significantly lower than controls, proposed lower endogenous pain control. No differences boys to girls chronic pain task; Parents of FAP group had greater pain-related fear, catastrophising, avoidance, helplessness, rumination (ρ < 0.001) and magnification (ρ < 0.012) about their child's pain. Children in FAP group significantly more likely to have a parent with chronic pain history.
Pate et al. (2019)	Qualitative semistructured interviews Community sample Australia	8 chronic pain, 8 healthy peers 8–12 years Mdn [IQR] = 10 [8.8–11.0] years	Chronic pain group, 1–2 years (63%) 3–5 years (25%) 6–10 years (13%)	8–12 years a Age analysis	Social constructs shown to have large impact on child's pain perception. Pain knowledge no different between groups. Less certainty with 8- and 9-year-old participants and less abstract understanding. Children focus on fixing the pain. Children with chronic pain more able to focus on the psychological, emotional and social impacts of pain.
Pavlova et al. (2020)	Cohort study Tertiary pain program past participants Western Canada	138 (75% female) 9–18 years M (SD) = 14.29 (2.30) years Mixed chronic pain conditions	M (SD) = 39.17 (38.66) months	ON	Older age poorer sleep, insomnia and shorter sleep duration. Older age greater pain interference and PTS. PTS associated with increased pain interference over time, decreased duration of sleep. Multiple pain areas in 47.2%. PTS greatest driver of pain interference. Worsening pain interference due to poor sleep quality
Pavone et al. (2011)	Case series, Outpatient pain clinic Italy	30 (12 female) 3–14 years $M=8$ years Growing pains	≥ 3 months	0 N	Massaging pain site mostly relieved pain. Family history of growing pains in 20%. Bilateral symptoms in 80% and linked to physical activity in 20%.
Pearson et al. (2011)	Case report Primary case Not stated	Single case report 8-year-old female CRPS	6–12 weeks	O N	Periods of excessive emotional vulnerability. Parental anxiety increased, stepfather to be deployed on overseas duty.
Perquin et al. (2000)	Cross sectional prevalence study Community sample Netherlands	5423 questionnaires completed and returned 0–18-year-olds	Prevalence study	Responses by age group categorised	25.0% reported chronic pain most commonly in age 12–15 years. Prevalence and multiple pain sites increased with age. Children up to 8 years, FAP was most common pain. Half of children with chronic pain reported more than one location of pain.

Author (date)	Methodology Participant recruitment Country	Participants N (sex) Age Diagnosis	Pain duration	Developmental framework	Primary findings
Seshia et al. (2008)	Cohort study Outpatient tertiary pain clinic Canada	70 (48 females, 22 males) 6–18 years M = 12 years (male) M = 14 years (female) Headaches	3–144 (<i>M</i> = 36) months	ON	Sleep disruption 23%; Multiple environment stressors reported by 46%; Isolation 7%; School stress 10%; Family history of recurrent chronic headaches 70%, anxiety 7%, mood disorder 31%.
Sieberg et al. (2011)	Cross sectional study Outpatient pain clinic USA	157 (87% female) 8–17 years M (SD) = 12.7 (2.43) years Multiple chronic pain conditions	M (SD) = 25.92 (13.79) months	ON	Parental factors: 30% reported significant global distress. Significant protective behaviours reported. Parental global stress, depression or anxiety did not directly influence level of child chronic pain disability, only parental pain related stress.
Sinclair et al. (2020)	Cross sectional study Outpatient pain clinic Australia	75 adolescents (68 girls, 7 boys) 13–18 years 39 children (30 girls, 9 boys) 8–12 years Multiple chronic pain conditions	≥ 3 months or CRPS criteria	Analysis of children and adolescents separately	Sensory modulation associated with attachment low registration linked to anxious and avoidant attachment. Higher sensory sensitivity related to lower anxious attachment and higher functional disability.
Soltani et al. (2020)	Case control study Outpatient pain clinic Canada	155 102 (71% girls) M (5D) = 14.20 (2.29) years Chronic pain group 53 controls (50% girls) 10–18 years M (5D) = 13.49 (2.71) years Mixed chronic pain conditions	M (5D) = 3.38 (3.25) years	ON	Initial and sustained attentional bias for high-level pain faces, high in both groups; whereas, chronic pain group gave more attention to low pain faces. Eye fixation measured on pain faces; fixation described as attentional bias to pain.
Tan et al. (2008)	Cross sectional study Outpatient pain clinic Netherlands	78 children (85.9% female) 5–16 years <i>M</i> = 13 years 951 adults CRPS type 1	<i>M</i> = 11.9 weeks	ON	Time from injury to symptoms 0.57 weeks (longer than adults). Children specific results: 23.3% one upper extremity, 72.6% lower extremity, and both extremities 4.1%. 28.2% had renewed CRPS type 1 and many due to a new injury, 60% in the same extremity. CRPS type 1 more likely to develop after minor injury in comparison to adults.

mental Primary findings ork	Poor pain perception at initial consult predicted treatment failure in primary care. Anxiety/depression did not predict treatment failure but emotional burden did. 23.3% of cohort reported disabling chronic pain at 3 months and continued to 6 months. Poor long-term prognosis is poor response to treatment 3 months in primary care. Older children at greater risk for persistent pain. Half of mothers and a third of fathers reported chronic	Reduced self-worth and low perceived academic performance increased symptom maintenance in combination with activity restriction. Punishment or pain disregard increased symptom maintenance. Activity avoidance not directly linked to pain intensity.	Low pain acceptance, catastrophising, and depression predictive of functional disability and less goal orientated behaviours.	Shift in resting state circuits from sensory alterations in paediatric cases with widespread brain variations, including issues with sensory processing and descending pain inhibition. Grey matter atrophy in right anterior cingulate cortex region and thalamus connections.	Attentional bias to painful and potentially harmful stimuli. Increased pain intensity at shorter latency in migraine group.
Developmental framework	0 2	0 N	0	0 Z	O Z
Pain duration	54.4 weeks	≥ 3 months	3–144 months M (<i>SD</i>) = 37 (28) months	M = 15 months	M(SD) = 5.7(1.7) years
Participants N (sex) Age Diagnosis	266 6–17 years M (SD) =11.2 (3.1) years Prospective of persistent pain	151 8–18 years (57% female) M (SD) = 12.1 (2.5) years Chronic abdominal pain	112 (76% female) 11–18 years M (SD) = 15.47 (1.83) years Mixed chronic pain conditions	52 and 52 controls Paediatric chronic cohort 16 (10 females, 6 males M (SD) = 14.3 years CRPS	16 <i>M</i> (<i>SD</i>) = 12 (1.5) years Chronic migraine Controls <i>M</i> (<i>SD</i>) = 12.3 (1.5) years 10–14 years
Methodology Participant recruitment Country	Prospective cohort study Primary care Germany	Cohort study Tertiary pain clinic USA	Cohort study Outpatient pain clinic USA	Case control study Outpatient tertiary pain clinic USA	Case control study Tertiary paediatric pain clinic and tertiary services Germany
Author (date)	Wager et al. (2019)	Walker et al. (2002)	Weiss et al. (2013)	Youssef et al. (2019)	Zohsel et al. (2008)

Note. BOLD = blood-oxygen-level-dependent; CRPS = complex regional pain syndrome; FAP = functional abdominal pain; JPFMS = juvenile primary fibromyalgia syndrome; ICD = International Classification of Diseases; PTS = post-traumatic stress; QoL = quality of life.

^a Explained that 8–12 years inclusion criterion was for developmental reasons.

Figure 2

Integrated Potential Influences on Onset and Early Trajectory of Childhood Chronic Pain

Context of child development

High levels of family support, protective behaviours or family disregard (Birnie, 2020; Dell'Api et al., 2007; Walker et al., 2002)

Parent diagnosis orientation & pain acceptance (Noel, Beals-Erickson et al., 2016)

Parent pain catastrophising (Akbarzadeh, 2018; Sieberg et al., 2011)

Child pain catastrophising (Libby & Glenwick; Weiss et al., 2013) Child anxiety (Bayle-Iniguez et al., 2015, Conte et al., 2003; Kashikar-Zuck et al., 2008)

Child age (Birnie 2020; Dell'Api, 2007; Perquin, 2000; Pate, 2019) Child sleep (Kanstrup et al., 2014; Konijnenberg et al., 2005; Pavlova et al., 2020)

Child stress (Bayle-Iniguez et al., 2015)

Child fear-avoidance and pain-related fear (Beekman et al., 2020; Dell'Api, 2007; Flack et al., 2017; Heathcote et al., 2017; Pas et al., 2019)

Potential influences on onset and early trajectory of childhood chronic pain

Child depression (Conte et al., 2003; Dunn et al., 2011; Kaczynski et al., 2009; Kanstrup et al., 2014; Libby & Glenwick, 2010; Weiss et al., 2013)

Child post-traumatic stress (Bayle-Iniguez et al., 2014; Pavlova et al., 2020; Pearson et al., 2011) Child psychological flexibility & self-efficacy (Beekman et al., 2020; Libby & Glenwick 2010)

Child pain attention (Soltani et al., 2020; Zohsel et al, 2008) Child neurobiological influences (Erpelding et al., 2014; Lebel et al., 2008; Linnman et al., 2013; Youssef et al., 2019)

Parent chronic pain (Libby, 2010; Pas, 2019; Seshia, 2008; Wager, 2019)

Parent anxiety or depression (Conte, 2003; Seshia, 2008)

Parent physical health (Birnie, 2020; Libby, 2010; Pas, 2019; Seshia 2008; Wager, 2019) Parent pain related stress (Abu-Arafeh & Abu-Arafeh; 2016, Carter, 2002; Sieberg et al., 2011)

Key:

Child-based influences.

Social influences.

measure (Huguet & Miró, 2008). However, increased age was associated with greater prevalence (Huguet & Miró, 2008) and pain interference (Birnie et al., 2020; Perquin et al., 2000). Neuropsychological developmental theory suggests that throughout childhood and adolescence, there are progressions in the ability to think and reason in abstract, in the development of insight into ones' own feelings, and in development of self-regulation skills (Anderson, 2002; Best & Miller, 2010). It

is unclear how such neuropsychological development relates to increased pain interference with age. However, consistent with such theory, in a qualitative study that took account of children's ages, younger children had less understanding of the abstract concept of pain and its purpose, and many children had difficulty communicating about the emotional aspects of pain (Pate et al., 2019).

Child neuro-biological influences

Neuroplasticity is the dynamic, neural variation in cortical networks. The cortical changes associated with neuroplasticity are related to adaptive learning (Fine & Sung, 2014). Studies of neurobiological circuits in children with complex regional pain syndrome (CRPS) utilising functional magnetic resonance imaging reported evidence of cortical reorganisation (Erpelding et al., 2014; Lebel et al., 2008; Linnman et al., 2013; Youssef et al., 2019) and grey matter atrophy (Youssef et al., 2019) that was concurrent with CRPS symptoms. Specifically, bloodoxygen-level-dependent (BOLD) signal alterations were found in areas representing descending pain inhibition, conditioned fear, and associated sensory processing regions (Erpelding et al., 2014; Lebel et al., 2008; Linnman et al., 2013). Youssef et al. (2019) meanwhile report prevalent grey matter atrophy in general sensory-emotional, motor, cognitive, and sensorydescending modulating brain regions, and describe a high degree of plastic cortical reorganisation in the thalamus connections in this population.

BOLD signal increases were also reported between the primary somatosensory cortex and the thalamus both in symptomatic and recovered CRPS groups (Lebel et al., 2008; Linnman et al., 2013). Mechanisms proposed for this persisting increase in connectivity in the children included learning processes from the trauma of CRPS symptoms and ongoing nociceptive recognition (Linnman et al., 2013). Concerningly, no known studies in children have ascertained the period over which these connective changes commence or resolve (Lebel et al., 2008; Linnman et al., 2013).

Child psychological factors exerting negative influences

Psychological factors play a key role influencing childhood chronic pain. Affective pain behaviours and the emotional aspects of pain experience were the strongest predictors of early treatment failure for children (Wager et al., 2019). Various baseline affective symptoms identified in this review that negatively affected outcomes included depression (Conte et al., 2003; Dunn et al., 2011; Kaczynski et al., 2009; Kanstrup et al., 2014; Libby & Glenwick, 2010; Weiss et al., 2013), anxiety (Bayle-Iniguez et al., 2015; Conte et al., 2003; Kashikar-Zuck et al., 2008), stress (Bayle-Iniguez et al., 2015); somatisation (Dunn et al., 2011), catastrophising (Libby & Glenwick, 2010; Weiss et al., 2013), pain-related fear (Flack et al., 2017; Heathcote et al., 2017; Pas et al., 2019), and fear-avoidance (Beeckman et al., 2020).

Depression or mood-related conditions, attention issues, greater behaviour-related problems, somatic symptoms, and anxiety were reported as pre-existing factors associated with chronic pain (Conte et al., 2003; Kashikar-Zuck et al., 2008). Anxiety was also associated with increased allodynia symptoms (de Tommaso et al., 2017). Children with chronic pain showed higher than average school functioning (Bayle-Iniguez et al., 2015; Ho et al., 2009), but along with this had perfectionist tendencies, general anxiety, and increased psychosocial stress (Bayle-Iniguez et al., 2015).

The review findings highlighted some variability, particularly in relation to children with a CRPS diagnosis. Retrospectively, CRPS participants self-reported fewer anxiety and depression

symptoms (Abu-Arafeh and Abu-Arafeh, 2016; Logan et al., 2013). However, it is plausible that the fewer psychological symptoms were primarily due to lower average pain duration and chronicity of symptoms of children in these two studies.

It is a common human process for pain to capture attention above other emotions (Soltani et al., 2020). Qualitative evidence reports that younger children interpreted their unexplained pain as life threatening (Dell'Api et al., 2007), with children's interpretation of their pain creating protective avoidant behaviours (Flack et al., 2017; Heathcote et al., 2017). Children with chronic pain were also more perceptive to all aspects of pain display, showing difficulty realigning their attention away from a painful sensation (Zohsel et al., 2008), described as an attentional bias (Soltani et al., 2020). Children's pain avoidance and attention bias was also described as anticipatory to the potential of pain, and not typically responsive to the sensation itself (Flack et al., 2017).

It is possible that avoidance behaviour in children with chronic pain is acquired via social or sociocultural learning. Consistently, neither activity avoidance nor participation levels were directly linked to pain intensity or symptoms (Beeckman et al., 2020; Walker et al., 2002). Instead, pain-related fear was the most central factor of activity avoidance and overall function (Beeckman et al., 2020). However, children with chronic pain who rated their self-worth poorly, similarly responded with greater functional disability and activity limitations, especially with activities they expected themselves to perform poorly in (Walker et al., 2002). This concept of low self-worth may be related to self-efficacy (Bandura, 2003).

Children's sleep

Children with chronic pain were shown to wake more frequently (Konijnenberg et al., 2005; Pavlova et al., 2020), with insomnia more common in older children and adolescents (Kanstrup et al., 2014; Pavlova et al., 2020). Insomnia explained the variance between chronic pain and functional disability, pain interference, and depression symptoms (Kanstrup et al., 2014; Pavlova et al., 2020). Higher ratings of baseline pain interference influenced greater sleep disturbances, and poor sleep quality was considered a major factor of pain symptom maintenance (Pavlova et al., 2020).

Child trauma and stress

Traumatic experiences can influence children's pain and stress response systems (Turner-Cobb, 2014). Various stressors were identified as preceding CRPS diagnosis (Bayle-Iniquez et al., 2015; Pearson & Bailey, 2011). The combined occurrence of poor-quality sleep and post-traumatic stress (PTS) are linked to chronic pain (Pavlova et al., 2020). Additionally, PTS correlated with increased pain intensity, pain impact, reduced quality of life (Noel, Wilson et al., 2016), and poor sleep (Pavlova et al., 2020). Further, both children with chronic pain and their parents presented with high levels of PTS (Noel, Wilson et al., 2016; Pavlova et al., 2020). This highlights the relevance of social learning models, whereby modelling or family influences may shape children's stress levels, and behavioural responses to pain and adversity. Sustained trauma is likely to influence children's long-term stress responses and emotional regulation (Davies, 2011; Steck & Steck, 2016), lending insight into the way PTS

might be associated with the onset and early trajectory of chronic pain.

Child psychological factors exerting positive influences

While many psychological factors are associated with the development of chronic pain, there are also psychological factors that positively influence chronic pain. Psychological flexibility is a personal value of adaptability and resilience and was found to govern activity participation more than pain itself (Beeckman et al., 2020). Psychological flexibility was also found to positively affect daily activity participation for older children with chronic pain through fewer fear-avoidance behaviours, by shifting cognitive attention to something else. Additionally, psychological flexibility was a mediator of goal orientated behaviours, decreased depression, and reduced functional disability (Beeckman et al., 2020). Psychological flexibility is reflective of neuropsychological development of executive function skills, where children gain increasing control over their ability to maintain and shift their attention (Best & Miller, 2010). Developmentally, this would not be as evident in younger children as it is a skill that develops with neuropsychological maturity (Best & Miller, 2010).

Self-efficacy, as theorised within SCT (Bandura, 2003) is another key resilience component of chronic pain and was a positive psychological factor highlighted by Libby and Glenwick (2010). Pain-related self-efficacy was positively associated with health-related quality of life measures in child chronic pain and was an identified moderator of depression and daily stress, and predicted pain ratings (Libby & Glenwick, 2010).

Social influences

Participation and peer interaction is an integral part of childhood developmental learning (Hoffnung, 2019), but unfortunately, the social consequences of reduced participation by children with chronic pain was highlighted in this review. Children with chronic pain had frequent school absenteeism (Kanstrup et al., 2014; Kashikar-Zuck et al., 2010). School absences were reported to be enabled by families, reinforcing illness behaviours, and creating positive attention for children's pain, thereby encouraging symptom maintenance (Walker et al., 2002).

Higher perceived support from family increased pain intensity (Libby & Glenwick, 2010), but negative support, in the form of disregard, also increased pain symptoms (Walker et al., 2002). Both findings can be explained by SCT, whereby incentive or disregard may prompt behaviour such as maintenance of symptom presentation (Miller, 2016).

Children who interacted more with healthcare professionals while continuing to seek resolution for their pain were reported to have poorer physical health and overall health satisfaction (Dell'Api et al., 2007). Notably, 40% of parents also had unresolved diagnosis orientation or a perception of incorrect diagnosis pertaining to their child's chronic pain symptoms (Noel, Beals-Erickson et al., 2016). From a SCT perspective, children observe and model their parents' behaviour (Miller, 2016). It is possible parents may reinforce children's low pain-acceptance behaviours (Weiss et al., 2013) through positive attention (Bandura, 2003), permitting withdrawal from unfavourable activities (Walker et al., 2002) or through the child

observing the parent's low pain acceptance beliefs regarding the child's pain (Noel, Beals-Erickson et al., 2016).

Consistent with developmental learning theories, parents were central mediators to childhood pain behaviours (Akbarzadeh et al., 2018). Children rely more strongly on their social support systems than themselves for pain related strategies and affirmations (Lynch et al., 2007). Vygotsky's SCDT posited that parents' interpretation of events determines a child's response to an event (Bergen, 2008). Aligning with SCDT tenets, several studies reported the concurrence of parental chronic pain or psychological mood disorders and their influence on their child's chronic pain onset (Conte et al., 2003; Kashikar-Zuck et al., 2010; Libby & Glenwick, 2010; Noel, Beals-Erickson et al., 2016; Pas et al., 2019; Seshia et al., 2008; Wager et al., 2019). Further, if a family member experienced chronic pain, children were more likely to catastrophise their own pain (Dell'Api et al., 2007). In 2020, Birnie et al. identified significant relationships between parent's physical and psychological health, protective behaviours, and child pain interference. Additional mediating factors such as parental pain catastrophising (Akbarzadeh et al., 2018), parental protective behaviours, and parent pain-related stress (Birnie et al., 2020; Sieberg et al., 2011) have also been shown to influence childhood chronic pain.

Higher levels of maternal pain catastrophising increased the mother's report of their child's pain intensity, anxiety, and depression. In contrast, high levels of a father's pain catastrophising only showed correlation to reports of their child's anxiety. Measures of mothers' catastrophising were significantly higher than those of fathers (Akbarzadeh et al., 2018). Drawing from SCT (Bandura, 1986, 2003) parental behaviours that relate to their own pain, or associated affective factors, are likely modelled, and could be readily assumed by their children (Steck & Steck, 2016).

Further adding to the complexity of these influences (see Figure 2), it appears that a child's pain impacts adversely on parent wellbeing (Carter, 2002; Hunfeld et al., 2002), which, in turn, mediates the child's functional ability (Sieberg et al., 2011). Parents did report increased stressors deriving from their child's pain (Abu-Arafeh & Abu-Arafeh, 2016); however, this stress was not present prior to the child's presenting pain complaint (Carter, 2002). A main stressor reported by these parents were the encounters with medical professionals, medical misinterpretations, and lack of diagnosis or treatment plans for their child's pain (Carter, 2002).

DISCUSSION

To our knowledge, this is the first study to integrate information about biological, psychological, and social influences with childhood developmental theories in relation to childhood chronic pain. The review findings provide an initial basis to inform practice for physiotherapists working with younger children with pain conditions in PC.

The literature in the review suggested that diverse biopsychosocial aspects including age at onset and pain duration, neuro-biological factors, psychological factors, parental impacts, sleep quality, trauma history, and stress interact with children's learning and development to collectively influence chronic pain onset and

early trajectory in children. Although the biopsychosocial model has served well as a basis for generating evidence about adult chronic pain, this review implies that, for children, the addition of a more nuanced, developmentally informed view of the influences may be appropriate. SCDT and SCT suggest children's learning about pain develops through observation and appraisal of situations, emotions, social interactions (Bandura, 2003; Miller, 2016; Smith, 2013), problem solving, and support from others (Koenig & Sabbagh, 2013; Miller, 2016; Vygotsky et al., 1978). From this learning comes complex and increasingly advanced abstract thinking about concepts such as pain, and insight into one's own and others' behaviours, including those relating to pain (Miller, 2016; Smith, 2013). While the review findings indicate developmental factors likely shape children's experiences of, and responses to, a pain event, children's early pain experiences also have potential to interrupt their development, through their impact on participation in childhood activities (Kanstrup et al., 2014; Kashikar-Zuck et al., 2010; Walker et al., 2002).

Implications for physiotherapy practice

The prognosis of chronic pain is thought to be determined from initial treatment effectiveness (Simons et al., 2018; Wager et al., 2019; Zernikow et al., 2018), where PC is the most common initial point of contact (Mallen et al., 2007). Physiotherapists working in PC are, therefore, ideally placed to provide quality, developmentally informed services early in a child's presentation where there are concerns over continuing pain, thereby interrupting the progression of the condition.

The findings from this review suggest that PC practitioners, but particularly physiotherapists, should understand that pain is experienced, not only through the physical realm, but also psychologically and socially, with all aspects needing to be appropriately managed for successful outcomes (Gatchel et al., 2007), as well as careful consideration given to children's development and factors that affect their learning. Furthermore, the influences of parental health, catastrophising, and protective or pain related behaviours need consideration (Birnie et al., 2020; Sieberg et al., 2011). To ensure a comprehensive and holistic evaluation of child and family factors in the early stages of a child's pain trajectory, an interprofessional approach is indicated. Physiotherapy service delivery in the PC setting should, therefore, be complemented by services provided by other professionals such as a paediatric psychologist and/or occupational therapist.

Since pain is a personal experience (Hinton & Kirk, 2016), and given the child influences identified in this review, it should be the child who is addressed in a consultation, while acknowledging the significant social support and information from parents. Recognising younger children's difficulty with understanding the abstract concept of pain (Pate et al., 2019), consultations should also draw on the opportunity to educate the child and their relevant social network, at cognitively appropriate levels, about the factors that can influence the child's pain experience and strategies that can be used. Parents may benefit from support in their interactions with their child (Noel, Beals-Erickson, et al., 2016).

From a learning perspective, it is possible that pain catastrophising (Dell'Api et al., 2007; Libby & Glenwick,

2010; Weiss et al., 2013) and fear-avoidance behaviours (Beeckman et al., 2020; Flack et al., 2017; Heathcote et al., 2017) communicate children's uncertainty and reflect their prior observations of others in pain. In terms of effective practice for physiotherapists and other primary care practitioners, catastrophising and fear avoidance might usefully be reconceptualised as worry or anxiety and searching for reassurance and understanding from parents and healthcare professionals (Levy et al., 2007). Therefore, physiotherapists should seek to understand what fears are creating anxiety, catastrophising, or avoidance behaviours.

Neuroscientific research and fMRI have advanced the understanding of pain experience (Steck & Steck, 2016). These imaging advances have also shown the variations of cortical connectivity networks and grey matter adaptations in children with CRPS (Bhatt et al., 2020; Lebel et al., 2008; Linnman et al., 2013). It is, therefore, imperative that these neurological adaptations are understood by all practitioners providing care in the onset and early trajectory of chronic pain in children.

Consistent with SCDT and SCT, parents are central mediators of children's pain responses, suggesting that at least some of children's responses are learned as part of a process involving their social and socio-cultural contexts (Akbarzadeh et al., 2018; Birnie et al., 2020). A family history of chronic pain can influence childhood chronic pain (Kashikar-Zuck et al., 2010; McKillop & Banez, 2016; Pas et al., 2019; Wager et al., 2019). Parents may assist a child in learning about activities to avoid (Walker et al., 2002) or reinforce avoidant behaviours through being extra-attentive to their child's pain (Sinclair et al., 2020), inadvertently increasing the child's functional disability.

It is plausible that parental protective behaviours are an adaptive response to their child's suffering, and feelings of failure to fix their child's pain (Carter, 2002; Maciver et al., 2010; Vasey et al., 2019). It is essential, therefore, that parents are not blamed by practitioners for their child's pain (Maciver et al., 2010), and learning theories suggest that parent's responses offer a means of supporting children to learn more adaptive responses to pain early in the trajectory. Parent pain responses in terms of how to feel and respond to pain, and the potential of reinforcement of pain are delicate, but potentially modifiable factors (Evans et al., 2008) and important treatment considerations for physiotherapists.

Limitations of the review

The findings of this review are constrained by the quality of studies that were included. There were various methodological limitations present within a large portion of the studies. For example, cross-sectional methodology was used in many studies in this review, and such studies provide limited insights into changes in children's pain trajectories and developmental influences over time. Additionally, there were limitations in terms of the included studies' ability to represent the population. Analysis of the age-related data relied, for the most part, on broad age-group means, and many of the studies involved, largely, adolescent populations. Analysis of children's developmental cognitive skills, essential to learning, were only present in a small number of studies (Lynch et al., 2007; Pate et al., 2019), limiting knowledge about younger children and their

understandings about pain. Generalisability of study findings to younger children attending PC settings was also constrained by the studies' predominant involvement of tertiary and specialist pain centre settings with children who had longstanding durations of their pain.

Relating to such concerns, it is a limitation of the review that a systematic, critical appraisal of the studies was not conducted, a requirement that is increasingly recognised within integrative review methodology and process (Toronto & Remington, 2020). While critical analysis and integration of the research in light of developmental theory was the basis of this review, further systematic review of the methodological quality of literature in this field remains an important next step.

Future research

Research into childhood chronic pain remains in its infancy. There is limited research investigating pain interpretation or pain experience in relation to development, especially in pre-adolescent children. As a result of this review, childhood pain researchers need to embrace the complexity of childhood development to assist in establishing robust practice guidelines. A formal interplay between research and developmental theory is required to urgently advance our clinical knowledge in this area (Huguet et al., 2011).

Future studies should acknowledge the range of factors associated with the onset and early trajectory of childhood chronic pain. Research is needed to ascertain whether associated central cortical changes resolve, whether there are variances in psychological influences with increased chronicity, and to establish optimal timelines for intervention effectiveness. There is a need for longitudinal research in primary healthcare settings, commencing in the early stages of children's pain conditions.

CONCLUSION

Younger children's pain is encompassed within continually changing cognitive, physical, and affective developmental stages. Children's social and family systems add further complexity to their pain experience. Adopting a conceptual understanding of children's pain in relation to their development would generate evidence that is better placed to inform practice. From a primary healthcare perspective, it is imperative that physiotherapists have a thorough understanding of the biopsychosocial influences and relevant developmental understandings to positively influence or interrupt children's chronic pain trajectories.

KEY POINTS

- Physiotherapists play a key role in managing the onset and early trajectory of chronic pain in children with initial treatment effectiveness affecting childhood chronic pain outcomes.
- 2. Research suggests physical, psychological, and social factors interact to influence the early onset and trajectory of childhood chronic pain.
- 3. Parental health, catastrophising, and protective behaviours exert specific influences on a child's pain interference and should be assessed.

4. Theory suggests children's cognitive development and learning shape their responses to pain.

DISCLOSURES

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PERMISSIONS

None.

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CONTRIBUTIONS OF AUTHORS

Conceptualisation and methodology – AM and MJ; Formal analysis and investigation – AM and MJ; Data curation – AM; Writing – original draft preparation – AM; Writing – review and editing – MJ; Supervision – MJ.

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Appendix A

Table A1

Supporting Childhood Development Texts

Assessment and development of executive function (EF) during childhood (Anderson, 2002).

Bandura's social cognitive theory: An introduction (Bandura, 2003).

Human development: Traditional and contemporary theories (Bergen, 2008).

A developmental perspective on executive function (Best & Miller, 2010).

Child development: A practitioner's guide (Davies, 2011).

Handbook of resilience in children (Goldstein & Brooks, 2013).

Lifespan development (4th Australasian ed.) (Hoffnung, 2019).

Theories of developmental psychology (6th ed.) (Miller, 2016).

Theories of human development (2nd ed.) (Newman, 2016).

A clinician's quide to normal cognitive development in childhood (Sandberg & Spritz, 2010).

Understanding children and childhood: A New Zealand perspective (5th ed.) (Smith, 2013).

Brain and mind: Subjective experience and scientific objectivity (2nd ed.) (Steck & Steck, 2016).

Child health psychology: A biopsychosocial perspective (Turner-Cobb, 2014).