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Systematic Review

Systematic Review: Predisposing, Precipitating, Perpetuating, and Present Factors Predicting Anticipatory Distress to Painful Medical Procedures in Children

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Abstract

Objective To conduct a systematic review of the factors predicting anticipatory distress to painful medical procedures in children. **Methods** A systematic search was conducted to identify studies with factors related to anticipatory distress to painful medical procedures in children aged 0–18 years. The search retrieved 7,088 articles to review against inclusion criteria. A total of 77 studies were included in the review. **Results** Thirty-one factors were found to predict anticipatory distress to painful medical procedures in children. A narrative synthesis of the evidence was conducted, and a summary figure is presented. **Conclusions** Many factors were elucidated that contribute to the occurrence of anticipatory distress to painful medical procedures. The factors that appear to increase anticipatory distress are child psychopathology, difficult child temperament, parent distress promoting behaviors, parent situational distress, previous pain events, parent anticipation of distress, and parent anxious predisposition. Longitudinal and experimental research is needed to further elucidate these factors.

Key words: anxiety; children; infancy; pain; parents; systematic review.

Introduction

Healthy children experience frequent medical procedures such as immunization and blood draws (Public Health Agency of Canada, 2006). Many young children experience high levels of pain and distress during these procedures, and adequate pain management strategies are seldom used (Lisi, Campbell, Pillai Riddell, Garfield, & Greenberg, 2013). Many children also experience distress and anxiety before the procedure even begins (Blount, Sturges, & Powers, 1990). This is called anticipatory distress. Anticipatory distress has been identified as occurring as early as infancy. Newborn infants who have been exposed to several painful procedures can learn to anticipate pain and exhibit more intense pain responses (Taddio, Shah, Gilbert-MacLeod, & Katz, 2002). Anticipatory distress and fear of medical procedures have also been identified as concerns in preschool and school-aged children. One study found that 22% of 4–6-year-old children experience serious distress during the preparatory phase of an immunization (Jacobson et al., 2001). Another recent study found that more than half of children under the age of 8 years have needle fear (Taddio et al., 2012). This finding is particularly concerning as anticipatory distress has been associated with several negative sequelae (Bijttebier & Vertommen, 1998; Palermo & Drotar, 1996; Tsao et al., 2004; Wright, Yelland, Heathcote, Ng, &

Wright, 2009). These negative outcomes could lead to avoidance of painful medical procedures and reduced compliance with preventative medical care (Taddio et al., 2012). Despite the important implications of anticipatory distress to painful medical procedures for children, little empirical work has investigated the factors that lead to its development.

Several models in the developmental literature have outlined the pathways that lead to the development of maladaptive anxiety and anxiety-related problems (Cicchetti & Cohen, 1995; Rachman, 1977; Vasey & Dadds, 2001). Within the pediatric pain literature, some work has examined the preprocedural child factors that impact a child's pain response (Kleiber & McCarthy, 2006; Young, 2005); however, these models focus on pain responses rather than anxiety and anticipatory distress. Previous models share a common emphasis on the transactional and developmental nature of anxiety or fear over time and highlight the dynamic interaction between the individual child and his/her environment. The four "Ps" of case formulation (predisposing, precipitating, perpetuating, and protective factors) also provide a useful framework for organizing the factors that may contribute to the development of anticipatory distress (Barker, 1988; Carr, 1999; Winters, Hanson, & Stoyanova, 2007). Predisposing factors are those that put a child at risk of developing a problem (in this case, high anticipatory distress). These may include genetics, life events, or temperament. Precipitating factors refer to a specific event or trigger to the onset of the current problem. Perpetuating factors are those that maintain the problem once it has become established. Finally, protective factors are strengths of the child or reduce the severity of problems and promote healthy and adaptive functioning. Another "P" that can be relevant in case formulation are "present" factors, that is, those that are operating during the time of the event-eliciting distress. Present factors are relevant due to the emphasis on "procedure" or context in the literature. Additionally, factors that are considered protective can be collapsed within predisposing, perpetuating, and present factors.

The objective of this review is to summarize the findings of studies that examine factors that predict anticipatory distress to painful medical procedures in children. This systematic review is a qualitative synthesis and summarizes the findings from the search in a summary figure. The goal of the summary figure (Figure 2) is to provide an overview for researchers and clinicians of the current literature as well as highlight gaps in the literature. Based on the developmental psychopathology perspective, factors in this review were hypothesized to fall under the four Ps of case formulation: predisposing (e.g., genetics and temperament), precipitating (e.g., negative pain experiences), perpetuating (e.g., parent behavior, parent anxiety, child behavior, and child cognitions), and present factors (e.g., health care professional behavior). This review also evaluated the included studies for risk of bias and identified methodological limitations of current studies. Promising directions for future research in this area are outlined.

Method

Criteria for Considering Studies for This Review

Types of Studies

Studies examining factors that are related to or predict anticipatory distress (anxiety, fear, distress) to painful medical procedures that were published in peer-reviewed journals were considered for inclusion. Although the goal of the study was to examine anticipatory distress, not pain, pain studies that measured anticipatory distress, anxiety or fear were included in the review. Given the study of fear and anxiety is a bourgeoning area in the field of pediatric pain and the goal was not to summarize treatment efficacy, nonrandomized studies were included in this review and formed the preponderance of the literature base. Nonrandomized studies were included following guidelines of the Cochrane Collaboration that a systematic review should include the best available study designs with the least risk of bias (Reeves, Deeks, Higgins, & Wells, 2011; Higgins & Green, 2011). Randomized controlled trials were included when appropriate; however, the variables predicting anticipatory distress were the focus, not the treatment effect. Pharmacological (e.g., sedatives) and physical (e.g., needle type) predictors of anticipatory distress were not examined in this review. All studies were examined for potential sources of bias.

Types of Participants

To be considered in the review, the study had to examine a painful procedure in children from birth through 18 years of age. The study also had to measure anticipatory distress (including anxiety/fear rated before or after the procedure or in some cases pain scores *prior* to the application of pain) to a painful medical procedure or operation (laboratory pain tasks were excluded from the review). Exclusion criteria for studies were: no painful medical procedure, incorrect age (i.e., not children 0 to 18 years), and studies where no factor was analyzed for its relationship to anticipatory distress.

Types of Measures of Anticipatory Distress

Studies that used an objective behavioral measure, observer reported (e.g., parent, nurse, physician, and research assistant), or self-report measure of distress prior to a painful medical procedure or operation were included in this review. In addition to distress

prior to painful medical procedures, for the purpose of this review, the term anticipatory distress was operationalized to be an umbrella term that also included ratings of fear or anxiety about a procedure provided after the procedure or operation as well as a retrospective report of anxiety/fear about a procedure. In circumstances where more than one measure of anticipatory distress was provided, self-report measures of anxiety, fear, and distress were prioritized. Behavioral measures and observer report measures were used when self-report was not available or was not developmentally appropriate. Additionally, measures that were most specific to anxiety and fear were used. For example, using the State Trait Anxiety Inventory over a general distress measure. Measures of anticipatory distress most proximal to the painful medical procedure were used. For preoperative studies, ratings in holding areas or during induction were used rather than during separation from parents as not to confound fear and distress of the medical procedure with fear and distress from separation.

Search Methods for Identification of Studies

A review protocol was not registered for this review. A librarian from a tertiary hospital with specialized training in conducting systematic reviews conducted a systematic search in MEDLINE, EMBASE, and EBM Reviews—Cochrane Central Register of Controlled Trials and PsycINFO to include articles indexed as of November 20, 2013. Separate search strategies and terms were developed for each of the databases. Search results were limited to publication years (1946+) and age group (children 0–18 years). Search terms related to anticipatory distress, medical procedures, pain procedures, and children were systematically paired (see Supplementary Appendix 1). A manual database search was also conducted for new articles published after 2013 to update the search in November 2014. Prior meta-analyses and reference lists from identified studies were also reviewed. Authors of studies that could not be found were contacted.

Data Collection and Analysis

Selection of Studies

Three authors (N.R., R.P.R., A.T.) and the librarian from a tertiary hospital identified studies through database searching as described above, and duplicates were removed using reference management software (Endnote X7). Two review authors (N.M.R. and R.R.P.R.) initially screened 1,000 abstracts to pilot the initial search strategy. Five review authors (N.M.R., R.R.P.R., P.T., M.C., and M.K.) screened titles and abstracts of studies from the final database searches for inclusion in the review based on predetermined inclusion and exclusion criteria listed above. Figure 1 provides the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA)

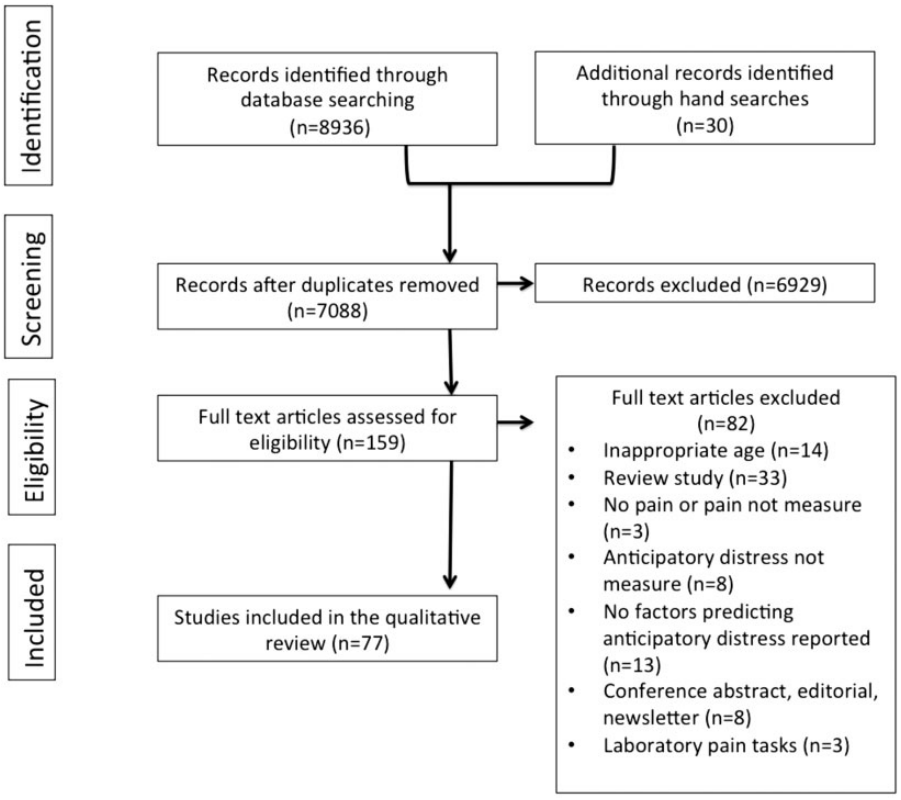

Figure 1. Included study flow chart following PRISMA guidelines.

(Moher, Liberati, Tetzlaff, & Altman, 2009) chart outlining the flow of study selection.

Data Extraction and Management

Four authors conducted data extraction independently for all included studies using a data extraction form created by the lead author designed for this review which was approved by the senior author (R.R.P.R.). The lead author conducted training sessions with the review authors to explicitly outline the exclusion criteria and how to use the data extraction form. Decision-making reliability for study inclusion was evaluated for 20% of all studies screened. Percent agreement, calculated as the percentage of studies that were agreed upon between two authors, ranged from 0.83 to 0.95 indicating strong inter-rater agreement.

Assessment of Risk of Bias

A nuanced approach was necessary as the purpose of this review was not to evaluate treatment outcomes or to make recommendations about practice. The state of the literature in the area of anticipatory distress is such that the preponderance of research is observational, not experimental, in nature. However, assessment of risk of bias within observational studies was deemed necessary despite the lack of randomization. Risk of bias was assessed for the 77 included studies using the Cochrane Collaboration methodology for systematic reviews (Higgins & Green, 2011). The majority of the studies included in the review (70 studies) were not randomized controlled trials. In the Risk of Bias tool created by the Cochrane Collaboration, the first three criteria (random sequence generation, allocation concealment, and blinding of participants) are only relevant for randomized controlled trials. As such, for observational and retrospective studies, only the last four criteria were used to make judgments: blinding of outcome assessment, incomplete outcome data, selective reporting, and other sources of bias. This adaptation was based on the decision not to penalize nonrandomized observational studies for being evaluated against criteria for randomization. The Cochrane collaboration recently launched a risk of bias tool for nonrandomized studies of interventions (Sterne, Higgins, & Reeves, 2014); however, at the time of this manuscript, trainings were only beginning to be offered and the tool was not yet widely used. Established tools to evaluate risk of bias in nonrandomized studies were also considered (e.g., Down & Black, 1998). However, given the number of studies in the review, an abbreviated tool was selected. Two authors evaluated risk of bias and consensus decisions were made where authors disagreed. All studies were classified as high, unclear, or low risk of bias. If one of the criteria was rated as "high," the overall study rating was considered to be high risk. "Unclear risk of bias" was indicated when one of the four criteria was missing, not mentioned, or did not meet the criteria for low or high risk of bias. To be evaluated as low risk of bias, all the criteria had to be rated as low.

Data Synthesis

Because of the diversity of medical procedures, outcome measures used, and participant ages included in the studies, a meta-analysis was not deemed appropriate for this review and, rather, a narrative synthesis framework (Popay et al., 2005) was applied. Influenced by developmental psychopathology theory and by the four Ps for case conceptualization (Vasey & Dadds, 2001; Winters, Hanson, & Stoyanova, 2007), this review categorized factors related to anticipatory distress to painful medical procedures as predisposing, precipitating, perpetuating, or present factors. Present factors were chosen (instead of protective factors) due to the emphasis on "procedure" or contextual factors in the literature. Additionally, factors that could be considered protective factors were collapsed within predisposing, perpetuating, and present factors as it made more conceptual sense based on how these factors were operationalized in the medical literature. For the purposes of this review, predisposing factors were operationalized as inherent variables that increase the child's risk for anticipatory distress, for example, preexisting aspects of the child such as age, gender, or temperament as well as sociodemographic variables of the parent or environment. Precipitating factors were conceptualized as factors that lead to the onset of anticipatory distress to painful medical procedures such as a negative pain event or previous experience with pain. Perpetuating or maintaining factors (Carr, 1999) were factors that likely extend or preserve the problem such as parent behavior that maintains the child's distress both inside and outside the medical procedure. Finally, present factors were variables that occurred at the time of the procedure and could positively or negatively influence the child's anticipatory distress. It should be noted that predisposing, precipitating, perpetuating, and present factors are not mutually exclusive categories as some factors may apply to multiple categories. These factors may also interact to compound anticipatory distress. For the purposes of the review, the lead author and senior author categorized each factor for parsimony and ease of interpretation.

Results

Results of the Search

The search strategy retrieved 7,088 abstracts to review against the inclusion criteria. Four individuals screened the initial 7,088 abstracts against inclusion criteria. Based on these criteria, the full article was

retrieved for 159 studies. Eighty-two articles for which the full text was retrieved were excluded from the review. A total of 77 full-text studies were included in the review. The review process followed the ~~Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA)~~ guidelines (Moher et al., 2009; Figure 1). Tables I–IV provide detailed overviews of the included studies including age range, sample size, country of origin, procedure, design, and risk of bias rating. In summary, most included studies were observational, from North America, encompassed a broad age range, and were based on surgical or needle-related procedures. The most common procedures included surgery or operative procedures (29), immunizations/injections (13), dental procedures (11), and venipuncture procedures (8). A total of 15,106 participants were included in the review.

In terms of the types of outcome measures, 43 measured anxiety, 15 measured fear, 13 measured distress, and 6 measured baseline pain. The majority of outcome measures were self-report (35), while the remaining outcome measures were behavioral (11) or observer reported (31).

Risk of Bias

Risk of bias was assessed for the 77 studies included in the review using the Cochrane Collaboration methodology for systematic reviews. As all but 7 studies were nonrandomized controlled trials, 15 studies were evaluated to have high risk of bias, 16 studies had unclear risk of bias, and 46 studies had low risk of bias (Tables I–IV).

Factors Included in the Review

The list of factors that predict anticipatory distress to painful medical procedures can be found in the leftmost column of Tables I–IV. Overall, there were 31 factors that were examined for their relationship to predict anticipatory distress.

Overall Findings

The overall goal of the review was to synthesize the literature on factors that predict anticipatory distress to painful medical procedures. Following data extraction, the lead and senior authors synthesized the results into the summary figure. The summary figure (Figure 2) includes most of the information from Tables I to IV and highlights the contribution of predisposing, precipitating, perpetuating, and present factors influencing the child's anticipatory distress. Only factors with two studies or more that were similar in nature were included in Figure 2. Moreover, in the summary figure, factors were subcategorized according to child, parent, health care professional, and/or contextual domains. Finally, the risk of bias and overall findings synthesis have been presented in Tables I–IV as well as in Figure 2.

Predisposing Factors

Child

As seen in Table I, 10 variables were identified for child predisposing factors (Table I). Results suggest that the data regarding age were inconclusive with almost half the studies showing no age effect, and the majority of the other studies suggesting younger children have higher anticipatory distress. The overall risk of bias for age was unclear (Table I). For gender, although results were varied, most studies (20/26) reported there was no effect of gender on anticipatory distress in children, while six studies found that girls experienced more anticipatory distress than boys. The overall risk of bias for gender was unclear (Table I). Four studies examined the effect of race on anticipatory distress, with the majority suggesting no effect. The overall risk of bias was unclear (Table I). Birth order was not found to have an effect on child anticipatory distress as indicated by three studies. The effect of number of siblings and sibling order was investigated by three studies and was found to have no effect. Child illness and child intelligence were both investigated by one study and were both found to positively predict child anticipatory distress.

For child psychopathology, the overall findings (6/8) supported the positive relationship between preexisting child psychopathology and increased child anticipatory distress to painful medical procedures. The overall risk of bias rating was unclear (Table I). For child temperament, the overall findings (8/11 studies) support a positive relationship between difficult child temperament and increased child anticipatory distress. The overall risk of bias score for temperament was unclear (Table I). Finally, there were inconclusive results for child attachment from two low risk of bias studies. One study (Horton et al., 2015) indicated that infants with avoidant infant attachment had lower anticipatory distress where as another study (Lumley, Melamed, & Abeles, 1993) found no effect.

Parent

A total of 12 studies provided evidence for parent predisposing factors that are associated with child anticipatory distress to painful medical procedures. Results are found in Table I. Overall findings suggest that parent anxious predisposition, and pain experience or fear of pain were all associated with increased anticipatory distress. The results for parent education and coping style were found to be inconclusive.

Health Professional

No health professional factors were found under the predisposing domain.

Table I. Predisposing Factors of Anticipatory Anxiety

Study	Age (years)	*N*	Country	Procedure	Design	Risk of bias	Result	Summary of bias	Results summary
Child predisposing factors									
1. Age (43 studies; $N = 9{,}890$)									
Bevan et al. (1990)	2–10	134	Canada	Preop	E	Unclear	–	Unclear (25 low, 8 high, 10 unclear)	Inconclusive
Broome and Hellier (1987)	5–11	84	USA	Medical	R	Low	–		
Caldwell-Andrews, Kain, Mayes, Kerns, and Ng (2005)	2–12	289	USA	Preop	O	Unclear	–		
Carpenter (1992)	4–18	73	USA	Venipuncture	O	Low	–		
Chen, Craske, Katz, Schwartz, and Zeltzer (2000)	3–18	55	USA	LP	RCT	High	–		
Chorney & Kain (2009)	2–10	293	USA	Preop	O	Low	–		
Chorney, Torry, McLaren, Chen, and Kain (2009)	2–10	293	USA	Preop	O	Unclear	–		
Dahlquist, Power, Cox, and Fernbach (1994)	2–7 8–17	63	USA	BMA	O	Low	–		
Dahlquist et al. (2001)	5–15	45	USA	Intramuscular injection and LP	O	Low	–		
Dahlquist and Pendley (2005)	2.4–5.1	29	USA	Immunization	RCT	High	–		
Davidson et al. (2006)	3–12	1,250	Australia	Preop	O	Low	–		
Fukuchi et al. (2005)	2–12	78	Brazil	Preop	O	Unclear	–		
Hatava, Olsson, and Lagerkranser (2000)	2–10	160	Sweden	Preop	E	Unclear	–		
Holm-Knudsen, Carlin, and McKenzie (1998)	0–14	2,122	Australia	Preop	O	Unclear	–		
Hosey et al. (2006)	2–14	407	UK	Dental	O	Low	–		
Howe et al. (2011)	4.9–16.2	23	USA	Injection, Finger sticks	O	Unclear	–		
Kain, Mayes, Weisman, and Hofstadter (2000)	3–10	60	USA	Preop	O	High	–		
Lilley, Craig, and Grunau (1997)	0.17–1.5	75	Canada	Immunization	O	Low	–		
Mahoney, Ayers, and Seddon (2010)	7–16	50	UK	Venipuncture	O	Low	–		
Melamed, Meyer, Gee, and Soule (1993)	4–12	46	USA	Preop	O	Low	–		
Olak et al. (2013)	8–10	344	Estonia	Dental	R	Low	–		
Taddio et al. (2012)	6–17	1,024	Canada	Immunization	R	High	–		
Tyc et al. (2002)	2–7	80	USA	Radiation Therapy	O	Low	–		
Kain, Mayes, O'Connor, and Cicchetti (1996)	2–10	163	USA	Preop	O	Low	+		
Tickle et al. (2009)	5–9	799	UK	Dental	O	High	+		
Al-Jundi and Mahmood (2010)	2–12	118	Jordan	Dental	O	High	∅		
Bijttebier and Vertommen (1998)	2.75–12.75	47	Belgium	Venipuncture	O	High	∅		
Carr, Lemanek, and Armstrong (1998)	3–12	62	USA	Allergy skin testing	O	Low	∅		
Claar, Walker, and Smith (2002)	8–18	100	USA	EGD	O	Low	∅		
deVos et al. (2012)	M = 3.1	18	USA	Immunotherapy Injections	O	High	∅		
Field, Alpert, Vega-Lahr, Goldstein, and Perry (1988)	4–10	56	USA	Preop	O	Low	∅		

(continued)

Table I. Continued

Study	Age (years)	*N*	Country	Procedure	Design	Risk of bias	Result	Summary of bias	Results summary
Fox and Newton (2006)	5–17	38	UK	Dental	RCT	Low	∅		
Goodenough, Champion, Laubreaux, Tabah, and Kampel (1998)	3–17	117	Australia	Venipuncture	O	Unclear	∅		
Jacobson et al. (2001)	1–6	150	USA	Immunization	O	Low	∅		
Lumley, Melamed, and Abeles (1993)	4–10	50	USA	Preop	O	Low	∅		
Ortiz et al. (2014)	8–16	437	Mexico	Dental	O	Unclear	∅		
McMurtry, Noel, Chambers, and McGrath (2011)	5–10	100	Canada	Venipuncture	O	Low	∅		
Mekarski and Richardson (1997)	2.5–13	324	Canada	Dental	O	Low	∅		
Messeri, Caprilli, and Busoni (2004)	2–14	39	Italy	Preop	O	Low	∅		
Siaw, Stephens, and Holmes (1986)	3.5–12.8	30	USA	Preop	O	Unclear	∅		
Thompson (1994)	8–12	43	USA	Preop	O	Low	∅		
Wright, Stewart, and Finley (2010)	3–6	61	Canada	Preop	E	Low	∅		
Wright, Stewart, and Finley (2013)	3–6	61	Canada	Preop	O	Low	∅		
2. Gender (26 studies; $N = 6{,}483$)									
Al-Jundi and Mahmood (2010)	2–12	118	Jordan	Dental	O	High	∅	Unclear (18 low, 6 high, 2 unclear)	No effect
Bearden, Feinstein, and Cohen (2012)	3–5	90	USA	Immunization	O	Low	∅		
Bijttebier and Vertommen (1998)	2.75–12.75	47	Belgium	Venipuncture	O	High	∅		
Carr, Lemanek, and Armstrong (1998)	3–12	62	USA	Allergy test	O	Low	∅		
Colares, Franca, Ferreira, Amorim Filho, and Oliverira (2013)	5–12	970	Brazil	Dental	R	Low	∅		
Dahlquist et al. (2001)	5–15	45	USA	Intramuscular injection and LP	O	Low	∅		
Davidson et al. (2006)	3–12	1,250	Australia	Preop	O	Low	∅		
Fortier, Martin, MacLaren Chorney, Mayes, and Kain (2011)	11–18	59	USA	Preop	O	Low	∅		
Fox and Newton (2006)	5–17	38	UK	Dental	RCT	Low	∅		
Gazal and Mackie (2007)	2–12	201	UK	Dental	O	Low	∅		
Goodenough et al. (1998)	3–17	117	Australia	Venipuncture	O	Unclear	∅		
Hanas et al. (2002)	1–15	41	Sweden	Insulin injection	RCT	High	∅		
Horton et al. (2015)	1–1.5	130	Canada	Immunization	O	Low	∅		
Jacobson et al. (2001)	1–6	150	USA	Immunization	O	Low	∅		
Kain et al. (2000)	3–10	60	USA	Preop	O	High	∅		
Lumley, Melamed, and Abeles (1993)	4–10	50	USA	Preop	O	Low	∅		
Ortiz et al. (2014)	8–16	437	Mexico	Dental	O	Unclear	∅		
Thompson (1994)	8–12	43	USA	Preop	O	Low	∅		
Wright, Stewart, and Finley (2010)	3–6	61	Canada	Preop	E	Low	∅		
Wright, Stewart, and Finley (2013)	3–6	61	Canada	Preop	O	Low	∅		
Broome and Hellier (1987)	5–11	84	USA	Medical	O	Low	+Girls		
Logan and Rose (2004)	12–18	102	USA	Preop	O	Low	+Girls		
McMurtry et al. (2011)	5–10	100	Canada	Venipuncture	O	Low	+Girls		
Olak et al. (2013)	8–10	344	Estonia	Dental	R	Low	+Girls		
Taddio et al. (2012)	6–17	1,024	Canada	Immunization	R	High	+Girls		
Tickle et al. (2009)	5–9	799	UK	Dental	O	High	+Girls		

(continued)

Table I. Continued

Study	Age (years)	*N*	Country	Procedure	Design	Risk of bias	Result	Summary of bias	Results summary
3. Child psychopathology (8 studies; $N = 2{,}053$)									
Davidson et al. (2006)	3–12	1,250	Australia	Preop	O	Low	+	Unclear (6 low, 2 high)	Positively predicts anticipatory distress.
Ericsson, Wadsby, and Hultcrantz (2006)	5–15	92	Sweden	Preop	RCT	High	+		
Fortier et al. (2011)	11–18	59	USA	Preop	O	Low	+		
Hosey et al. (2006)	2–14	407	UK	Dental	O	Low	+		
Kain et al. (2000)	3–10	60	USA	Preop	O	High	+		
Wright, Stewart, and Finley (2013)	3–6	61	Canada	Preop	O	Low	+		
Kiley and Polillio (1997)	School age	74	USA	Immunization	E	Low	∅		
Lumley, Melamed, and Abeles (1993)	4–10	50	USA	Preop	O	Low	∅		
4. Temperament (11 studies; $N = 2{,}235$)									
Arnrup, Broberg, Berggren, and Bodin (2003)	4–12	86	Sweden	Dental	E	High	+	Unclear (7 low, 3 high, 1 unclear)	Positively predicts anticipatory distress.
Chen et al. (2000)	3–18	55	USA	LP	RCT	High	+		
Cropper et al. (2011)	4–7	84	UK	GA (Cochlear Implant)	O	Low	+		
Fortier et al. (2011)	11–18	59	USA	Preop	O	Low	+		
Jacobson et al. (2001)	1–6	150	USA	Immunization	O	Low	+		
Kain et al. (1996)	2–10	163	USA	Preop	O	Low	+		
Kain et al. (2000)	3–10	60	USA	Preop	O	High	+		
Lee and White-Traut (1996)	3–7	137	USA	Venipuncture	O	Unclear	+		
Davidson et al. (2006)	3–12	1,250	Australia	Preop	O	Low	∅		
Horton et al. (2015)	1–1.5	130	Canada	Immunization	O	Low	∅		
Wright, Stewart, and Finley (2013)	3–6	61	Canada	Preop	O	Low	∅		
5. Race (4 studies; $N = 296$)									
Broome and Hellier (1987)	3–15	140	USA	Medical	R	Low	∅	Unclear (3 low, 1 high)	No effect
Kain et al. (2000)	3–10	60	USA	Preop	O	High	∅		
Lumley, Melamed, and Abeles (1993)	4–10	50	USA	Preop	O	Low	∅		
Melamed et al. (1993)	4–12	46	USA	Preop	O	Low	+		
6. Birth order (3 studies; $N = 1{,}352$)									
Davidson et al. (2006)	3–12	1,250	Australia	Preop	O	Low	∅	Low (3 low)	No effect
Fortier et al. (2011)	11–18	59	USA	Preop	O	Low	∅		
Thompson (1994)	8–12	43	USA	Preop	O	Low	∅		
7. Number of siblings/sibling order (3 studies; $N = 1{,}369$)									
Davidson et al. (2006)	3–12	1,250	Australia	Preop	O	Low	∅	Unclear (2 low, 1 high)	No effect
Fortier et al. (2011)	11–18	59	USA	Preop	O	Low	∅		
Kain et al. (2000)	3–10	60	USA	Preop	O	High	∅		
8. Child illness (1 study; $N = 80$)									
Tyc et al. (2002)	2–7	80	USA	Radiation Therapy	O	Low	+	Low	CNS disease positively predicts.
9. Attachment (2 studies; $N = 180$)									
Horton et al. (2015)	1–1.5	130	Canada	Immunization	O	Low	−	Low	Inconclusive
Lumley, Melamed, and Abeles (1993)	4–10	50	USA	Preop	O	Low	∅		

(continued)

Table I. Continued

Study	Age (years)	*N*	Country	Procedure	Design	Risk of bias	Result	Summary of bias	Results summary
10. Intelligence (1 study; $N = 60$)									
Kain et al. (2000)	3–10	60	USA	Preop	O	High	+	High	Positively predicts anticipatory distress.
Parent predisposing factors									
1. Anxious predisposition (4 studies; $N = 1{,}532$)									
Davidson et al. (2006)	3–12	1,250	Australia	Preop	O	Low	+	Low	Positively predicts anticipatory distress.
Kain et al. (1996)	2–10	163	USA	Preop	O	Low	+		
Messeri, Caprilli, and Busoni (2004)	2–14	39	Italy	Preop	O	Low	+		
Tyc et al. (2002)	2–7	80	USA	Radiation Therapy	O	Low	∅		
2. Beliefs about coping and coping style (2 studies; $N = 349$)									
Caldwell-Andrews et al. (2005)	2–12	289	USA	Preop	O	Unclear	–	Unclear	Inconclusive.
Kain et al. (2000)	3–10	60	USA	Preop	O	High	+		
3. Pain experience and fear of pain (3 studies; $N = 1{,}185$)									
Ellerton and Merriam (1994)	3–15	75	Canada	Preop	R	Unclear	+	High (1 unclear, 2 high)	Positively predicts anticipatory distress.
Arnup (2003)	4–12	86	Sweden	Dental	E	High	∅		
Taddio et al. (2012)	6–17	1,024	Canada	Immunization	R	High	+		
4. Parental education (2 studies; $N = 1{,}029$)									
Colares et al. (2013)	5–12	970	Brazil	Dental	R	Low	+	Low (2 low)	Inconclusive
Fortier et al. (2011)	11–18	59	USA	Preop	O	Low	∅		
5. Parent gender (1 study; $N = 437$)									
Ortiz et al. (2014)	8–16	437	Mexico	Dental	O	Unclear	+	Unclear	Mothers associated with more anticipatory distress.
Contextual predisposing factors									
1. Previous hospitalization of child or sibling (5 studies; $N = 1{,}451$)									
Broome and Hellier (1987)	5–11	84	USA	Medical	R	Low	∅	Unclear (4 low, 1 high)	No effect of previous hospitalization.
Field et al. (1988)	4–10	56	USA	Preop	O	Low	∅		
deVos et al. (2012)	M = 3.1	18	USA	Immunotherapy Injections	O	High	∅		
Thompson (1994)	8–12	43	USA	Preop	O	Low	∅		
Broome and Hellier (1987)	5–11	84	USA	Medical	R	Low	+ (sibling)		
Davidson et al. (2006)	3–12	1,250	Australia	Preop	O	Low	+		
2. Other contextual factors (4 studies; $N = 3{,}079$)									
Colares et al. (2013)	5–12	970	Brazil	Dental	R	Low	+ (no visits)		

Table I. Continued

Study	Age (years)	*N*	Country	Procedure	Design	Risk of bias	Result	Summary of bias	Results summary
Davidson et al. (2006)	3–12	1,250	Australia	Preop	O	Low	∅ (SES)	Unclear (2 low, 2 high)	No/irregular dental visits positively predict.
Kain et al. (2000)	3–10	60	USA	Preop	O	High	∅(demographics)		
Tickle et al. (2009)	5–9	799	UK	Dental	O	High	+ (visits)		

Note. + Factor has a positive relationship with anticipatory distress; − Factor has a negative relationship with anticipatory distress; ∅ = no effect or significant relationship; O = observational study; E = experimental study; R = retrospective study; RCT = randomized controlled trial; Preop = preoperative; LP = lumbar puncture; BMA = bone marrow aspiration; CNS = central nervous system.

Contextual

Two factors were identified as contextual predisposing factors: previous hospitalization of the child or sibling (five studies) and other contextual factors (four studies). For previous hospitalization, four of the five studies found no effect of previous hospitalization on child anticipatory distress; however, one study (Broome & Hellier, 1987) found that hospitalization of a sibling (but not of self) was associated with higher anticipatory distress. The overall risk of bias for this factor was unclear. In terms of other contextual factors, the overall findings showed that having never visited a dentist and having irregular visits to the dentist are positively associated with child anticipatory distress. Other demographic variables had mixed results. The evidence for this factor was unclear.

Precipitating Factors

Child

Two broad factors were identified as precipitating factors that contributed to the onset of anticipatory distress to painful medical procedures (Table II). The first factor was general and specific negative pain events (33 studies). The results showed that 17 studies found that previous negative experiences positively predict child anticipatory distress, whereas 12 studies found no effect of previous painful events. Four studies indicated that history of painful procedures was in fact associated with decreased child anticipatory distress. Overall, the evidence points toward a positive relationship between previous pain events and child anticipatory distress based on unclear risk of bias.

The second precipitating child factor that was identified was previous child/adolescent behavior (five studies). Overall, results indicate that previous pain behavior positively predicts child anticipatory distress. The risk of bias was unclear.

Parent

No parent factors were found under the precipitating domain.

Health Professional

No health professional factors were found under the precipitating domain.

Contextual

No contextual factors were found under the precipitating domain.

Perpetuating Factors

Child

As listed in Table III, four factors were identified as child perpetuating factors: child knowledge (seven studies), child coping style (four studies), child cognitions (three studies), and other child behaviors (two

Table II. Precipitating Factors of Anticipatory Anxiety

Study	Age (years)	*N*	Country	Procedure	Design	Risk of bias	Result	Summary of bias	Results summary
Child precipitating factors									
1. General and specific negative pain events (33 studies; $N = 5{,}186$)									
Al-Jundi and Mahmood (2010)	2–12	118	Jordan	Dental	O	High	+	Unclear (20 low, 7 high, 6 unclear)	Positively predicts anticipatory distress.
Bijttebier and Vertommen (1998)	2.75–12.75	47	Belgium	Venipuncture	O	High	+		
Caes et al. (2014)	0.6–15	28	Canada	BMA or LP	O	Unclear	+		
Carillo-Diaz, Crego, Armfield, and Romero (2013)	8–18	179	Spain	Dental	R	Unclear	+		
Colares et al. (2013)	5–12	970	Brazil	Dental	R	Low	+		
Cropper et al. (2011)	4–7	84	UK	Preop	O	Low	+		
Ellerton and Merriam (1994)	3–15	75	Canada	Preop	R	Unclear	+		
Hatava, Olsson, and Lagerkranser (2000)	2–10	160	Sweden	Preop	E	Unclear	+		
Jacobson et al. (2001)	1–6	150	USA	Immunization	O	Low	+		
Kain et al. (1996)	2–10	163	USA	Preop	O	Low	+		
Lee and White-Traut (1996)	3–7	137	USA	Venipuncture	O	Unclear	+		
Lumley, Melamed, and Abeles (1993)	4–10	50	USA	Preop	O	Low	+ (quality)		
Noel, McMurtry, Chambers, and McGrath (2010)	5–10	48	Canada	Venipuncture	O	Low	+		
Olak et al. (2013)	8–10	344	Estonia	Dental	R	Low	+		
Pillai Riddell et al. (2011)	0–1	731	Canada	Immunization	O	Low	+		
Taddio et al. (2002)	Newborns (>1 month)	66	Canada	Venipuncture, Vitamin K injections	O	Low	+		
Tickle et al. (2009)	5–9	799	UK	Dental	O	High	+		
Arnrup et al. (2003)	4–12	86	Sweden	Dental	E	High	∅		
Broome, Lillis, McGahee, and Bates (1994)	3–15	14	USA	LP	O	Low	∅		
Carr, Lemanek, and Armstrong (1998)	3–12	62	USA	Allergy testing	O	Low	∅		
Chorney & Kain (2009)	2–10	293	USA	Preop	O	Low	∅		
Ericcson, Wadsby, and Hultcrantz (2006)	5–15	92	Sweden	Preop	RCT	High	∅		
Dahlquist et al. (2001)	5–15	45	USA	Intramuscular injection and LP	O	Low	∅		
Fortier et al. (2011)	11–18	59	USA	Preop	O	Low	∅		
Goubet, Clifton, and Shah (2001)	0–0.04	12	USA	Heel-lance	O	Low	∅		
Owens and Todt (1984)	Newborns (>1 month)	20	USA	Heel-lance	O	Low	∅		
McMurtry et al. (2011)	5–10	100	Canada	Venipuncture	O	Low	∅		
Wright, Stewart, and Finley (2010)	3–6	61	Canada	Preop	E	Low	∅		
Wright, Stewart, and Finley (2013)	3–6	61	Canada	Preop	E	Low	∅		
Mahoney, Ayers, and Seddon (2010)	7–16	50	UK	Venipuncture	O	Low	−		
deVos et al. (2012)	$M = 3.1$	18	USA	Immunotherapy Injections	O	High	−		

(continued)

Table II. Continued

Study	Age (years)	*N*	Country	Procedure	Design	Risk of bias	Result	Summary of bias	Results summary
Howe et al. (2011)	4.9–16.2	23	USA	Insulin injection and Finger sticks	O	Unclear	–		
Hanas et al. (2002)	1–15	41	Sweden	Insulin injection	RCT	High	–		
2. Previous pain behavior (5 studies; $N = 3,681$)								Unclear (4 low, 1 unclear)	Positively predicts anticipatory distress.
Holm-Knudsen, Carlin, and McKenzie (1998)	0–14	2,122	Australia	Preop	O	Unclear	+		
Davidson et al. (2006)	3–12	1,250	Australia	Preop	O	Low	+		
Jacobson et al. (2001)	1–6	150	USA	Immunization	O	Low	+		
McMurtry et al. (2011)	5–10	100	Canada	Venipuncture	O	Low	+		
Fortier et al. (2011)	11–18	59	USA	Preop	O	Low	∅		

Note. + Factor has a positive relationship with anticipatory distress; – Factor has a negative relationship with anticipatory distress; ∅, no effect or significant relationship; O = observational study; E = experimental study; R = retrospective study; RCT = randomized controlled trial; Preop = preoperative; LP = lumbar puncture; BMA = bone marrow aspiration.

studies). For child knowledge, the results were inconclusive. This was based on unclear evidence. The evidence for child coping style was inconclusive based on unclear risk of bias. For child cognitions (three studies), overall results suggest that child cognitions including high threat appraisal, lower perceived control, and high aversion to the procedure were all associated with higher child anticipatory distress, based on studies with unclear risk of bias. Finally, the evidence for other child behaviors (two studies) was inconclusive as studies highlighted different child behaviors associated with increased or decreased anticipatory distress.

Parent

Four factors were identified as parent perpetuating factors: parent behavior (7 studies), parent situational distress (19 studies), parent anticipation of child distress (5 studies), and parent self-efficacy/attitudes (2 studies) (Table III). Overall findings suggest that most parent behavior, parent situational distress, and parent anticipation of child distress were associated with increased anticipatory distress (Table III).

Health professional

No health professional factors were found under the perpetuating domain.

Contextual

No contextual factors were found under the perpetuating domain.

Present Factors

Child

As listed in Table IV, one factor was identified for child present factors: idiosyncratic needs. One study (Ameringer, Elswick Jr, Shockey, & Dillon, 2013) showed that fatigue and nausea were positively associated with child anticipatory distress prior to chemotherapy with a low risk of bias.

Parent

One parent present factor was identified: parental presence during a painful medical procedure. The overall results for this factor are inconclusive (Table IV). The risk of bias for this factor was unclear.

Health Professional

One factor was identified as a health professional present factor: health professional behavior (three studies). Overall, evidence suggests that distress promoting behavior by health care professionals is associated with higher child anticipatory distress. The overall risk of bias was unclear.

Table III. Perpetuating Factors of Anticipatory Anxiety

Study	Age (years)	*N*	Country	Procedure	Design	Risk of bias	Result	Summary of bias	Results summary
Child perpetuating factors									
1. Child knowledge (7 studies; $N = 1{,}850$)									
Claar, Walker, and Barnard (2002)	8–17	100	USA	EGD	O	Low	∅	Unclear (5 low, 2 unclear)	Inconclusive
Crandall, Lammers, Senders, Braun, and Savedra (2008)	7–13	60	USA	Preop	E	Low	∅		
Jacobson et al. (2001)	1–6	150	USA	Immunization	O	Low	∅		
Claar, Walker, and Smith (2002)	8–18	100	USA	EGD	O	Low	∅		
Davidson et al. (2006)	3–12	1,250	Australia	Preop	O	Low	−		
Hatava, Olsson, and Lagerkranser (2000)	2–10	160	Sweden	Preop	E	Unclear	−		
Siaw, Stephens, and Holmes (1986)	3.5–12.8	30	USA	Preop	O	Unclear	−		
2. Child coping style (4 studies; $N = 174$)									
Bijttebier and Vertommen (1998)	2.75–12.75	47	Belgium	Venipuncture	O	High	+	Unclear (1 high, 3 low)	Inconclusive
Field et al. (1988)	4–10	56	USA	Preop	O	Low	∅		
Smith, Ackerson, and Blotcky (1989)	6–18	28	USA	BMA and LP	O	Low	∅		
Thompson (1994)	8–12	43	USA	Preop	O	Low	−		
3. Other child behaviors (2 studies; $N = 368$)									
Chorney & Kain (2009)	2–10	293	USA	Preop	O	Low	+	Unclear (1 low, 1 unclear)	More research needed.
Kain et al. (1998)	2–12	75	USA	Preop	RCT	Unclear	−		
4. Child cognitions (3 studies; $N = 352$)									
Carillo-Diaz et al. (2013)	8–18	179	Spain	Dental	R	Unclear	+ (expectancy)	Unclear (2 low, 1 unclear)	Negative child cognitions positively predict child anticipatory distress.
Claar, Walker, and Smith (2002)	8–18	100	USA	EGD	O	Low	+		
Carpenter (1992)	4–18	73	USA	Venipuncture	O	Low	_		
Carillo-Diaz et al. (2013)	8–18	179	Spain	Dental	R	Unclear	+ (appraisal)		
Parent perpetuating factors									
1. Parent behavior (7 studies; $N = 1{,}962$)									
Blount, Sturges, and Powers (1990)	5–13	22	USA	BMA or LP	O	Unclear	+	Unclear (5 low, 2 unclear)	Parent behaviour is associated with anticipatory distress. Direction dependent on type of behavior.
Chorney et al. (2009)	2–10	293	USA	Preop	O	Unclear	+, − (distraction)		
Dahlquist Power, Cox, and Fernbach (1994)	2–7, 8–17	63	USA	BMA	O	Low	+		
Dahlquist et al. (2001)	5–15	45	USA	Intramuscular injection and LP	O	Low	+		
Lisi et al. (2013)	0–1	760	Canada	Immunization	O	Low	+		
Noel et al. (2010)	5–10	48	Canada	Venipuncture	O	Low	+		
Pillai Riddell et al. (2011)	0–1	731	Canada	Immunization	O	Low	∅		
2. Parent situational distress (19 studies; $N = 4{,}998$)									
Arnrup et al. (2003)	4–12	86	Sweden	Dental	E	High	+	Unclear (7 high, 9 low, 3 unclear)	Positively predicts anticipatory distress.
Bearden et al. (2012)	3–5	90	USA	Immunization	O	Low	+		
Bevan et al. (1990)	2–10	134	Canada	Preop	E	Unclear	+		
Caes et al. (2014)	0.6–15	28	Canada	BMA or LP	O	Unclear	+		
Colares et al. (2013)	5–12	970	Brazil	Dental	R	Low	+		
Dahlquist Power, Cox, and Fernbach (1994)	2–7, 8–17	63	USA	BMA	O	Low	+		
Davidson et al. (2006)	3–12	1,250	Australia	Preop	O	Low	+		
Gazal and Mackie (2007)	2–12	201	UK	Dental	O	Low	+		

(continued)

Table III. Continued

Study	Age (years)	*N*	Country	Procedure	Design	Risk of bias	Result	Summary of bias	Results summary
Hatava, Olsson, and Lagerkranser (2000)	2–10	160	Sweden	Preop	E	Unclear	+		
Kain et al. (2000)	3–10	60	USA	Preop	O	High	+		
LaMontagne, Hepworth, Johnson, and Cohen (1996)	8–17	90	USA	Preop	O	High	+		
Messeri, Caprilli, and Busoni (2004)	2–14	39	Italy	Preop	O	Low	+		
Olak et al. (2013)	8–10	344	Estonia	Dental	R	Low	+		
Tickle et al. (2009)	5–9	799	UK	Dental	O	High	+		
Tourigny (1992)	2–10	50	Canada	Preop	O	High	+		
Al-Jundi and Mahmood (2010)	2–12	118	Jordan	Dental	O	High	∅		
Dahlquist and Pendley (2005)	2.4–5.1	29	USA	Immunization	RCT	High	∅		
Hosey et al. (2006)	2–14	407	UK	Dental	O	Low	∅		
Tyc et al. (2002)	2–7	80	USA	Radiation Therapy	O	Low	∅		
3. Parent anticipation of child distress (5 studies; $N = 742$)									
Jacobson et al. (2001)	1–6	150	USA	Immunization	O	Low	+	Unclear (1 unclear, 4 low)	Positively predicts anticipatory distress.
Ortiz et al. (2014)	8–16	437	Mexico	Dental	O	Unclear	+		
Lumley, Melamed, and Abeles (1993)	4–10	50	USA	Preop	O	Low	+		
Tyc et al. (2002)	2–7	80	USA	Radiation Therapy	O	Low	+		
Srivastava, Betts, Rosenberg, and Kainer (2001)	0–6.5	25	Australia	Micturating cystoure throgram	O	Low	∅		
4. Parent self-efficacy/attitude toward procedure (2 studies; $N = 236$)									
Arnrup et al. (2003)	4–12	86	Sweden	Dental	E	High	∅	Unclear	More research is needed.
Jacobson et al. (2001)	1–6	150	USA	Immunization	O	Low	∅		

Note. + Factor has a positive relationship with anticipatory distress; − Factor has a negative relationship with anticipatory distress; ∅, no effect or significant relationship; O = observational study; E = experimental study; R = retrospective study; RCT = randomized controlled trial; Preop = preoperative; LP = lumbar puncture; BMA = bone marrow aspiration; EGD = esophagogastroduodenoscopy.

Table IV. Present Factors of Anticipatory Anxiety

Study	Age (years)	*N*	Country	Procedure	Design	Risk of bias	Result	Summary of bias	Results summary
Child present factors									
1. Idiosyncratic needs (1 study; $N = 9$)									
Ameringer et al. (2013)	13–18	9	USA	Chemotherapy	O	Low	+	Low	More research is needed.
Parent present factors									
1. Parent presence (6 studies; $N = 2{,}159$)									
Al-Jundi and Mahmood (2010)	2–12	118	Jordan	Dental	O	High	∅	Unclear (3 low, 2 high, 1 unclear)	Inconclusive
Bevan et al. (1990)	2–10	134	Canada	Preop	E	Unclear	∅		
Davidson et al. (2006)	3–12	1,250	Australia	Preop	O	Low	∅		
Tourigny (1992)	2–10	50	Canada	Preop	O	High	+		
Messeri, Caprilli, and Busoni (2004)	2–14	39	Italy	Preop	O	Low	−		
Kain et al. (2006)	2–12	568	USA	Preop	O	Low	−,+		
Health care professional factors									
1. Health professional behavior (3 studies; $N = 386$)									
Noel et al. (2010)	5–10	48	Canada	Venipuncture	O	Low	+	Unclear (2 low, 1 unclear)	Distress promoting behaviour positively predicts child anticipatory distress.
Chorney et al. (2009)	2–10	293	USA	Preop	O	Unclear	+		
Dahlquist et al. (2001)	5–15	45	USA	Intramuscular injection and LP	O	Low	+ (nurse) ∅ (MD)		
Contextual present factors									
1. Environmental factors (15 different studies; $N = 4{,}926$)									
Davidson et al. (2006)	3–12	1,250	Australia	Preop	O	Low	− admission type	Unclear (9 low, 4 high, 2 unclear)	More research needed.
Holm-Knudsen, Carlin, and McKenzie (1998)	0–14	2,122	Australia	Preop	O	Unclear	− induction location		
Kain, Wang, Mayes, Krivutza, and Teague (2001)	2–7	70	USA	Preop	RCT	High	− reduced sensory stimulation		
Mekarski and Richardson (1997)	2.5–13	324	Canada	Dental	O	Low	+ dental work severity		
deVos et al. (2012)	$M = 3.1$	18	USA	Immunotherapy Injections	O	High	∅ injection personnel		
Horton et al. (2015)	1–1.5	130	Canada	Immunization	O	Low	∅ number of needles		
Davidson et al. (2006)	3–12	1,250	Australia	Preop	O	Low	+ longer procedure		
deVos et al. (2012)	M = 3.1	18	USA	Immunotherapy Injections	O	High	+ intervals between needles		
Holm-Knudsen, Carlin, and McKenzie (1998)	0–14	2,122	Australia	Preop	O	Unclear	∅ fasting time		
Al-Jundi and Mahmood (2010)	2–12	118	Jordan	Dental	O	High	+ referral reason		
Dahlquist Power, Cox, and Fernbach (1994)	2–7, 8–17	63	USA	BMA	O	Low	∅ time since diagnosis		
Dahlquist et al. (2001)	5–15	45	USA	Intramuscular injection and LP	O	Low	∅ time since diagnosis		
Tyc et al. (2002)	2–7	80	USA	Radiation Therapy	O	Low	∅ time since diagnosis		
Dahlquist and Pendley (2005)	2.4–5.1	29	USA	Immunization	RCT	High	− time since diagnosis		
Holm-Knudsen, Carlin, and McKenzie (1998)	0–14	2,122	Australia	Preop	O	Unclear	∅ type of case		
Ortiz et al. (2014)	8–16	437	Mexico	Dental	O	Unclear	∅ procedure		
Wright, Stewart, and Finley (2010)	3–6	61	Canada	Preop	E	Low	∅ surgery type		
Wollin et al. (2004)	5–12	120	Australia	Preop	O	Low	+ various factors		

(continued)

Table IV. Continued

Study	Age (years)	N	Country	Procedure	Design	Risk of bias	Result	Summary of bias	Results summary
deVos et al. (2012)	$M = 3.1$	18	USA	Immunotherapy Injections	O	High	∅ number of needles		
Fortier et al. (2011)	11–18	59	USA	Preop	O	Low	∅ preadmission visit		

Note. +, factor has a positive relationship with anticipatory distress; −, factor has a negative relationship with anticipatory distress; ∅, no effect or significant relationship; O = observational study; E = experimental study; R = retrospective study; RCT = randomized controlled trial; Preop = preoperative; LP = lumbar puncture; BMA = bone marrow aspiration.

Contextual

One broad factor, environmental factors, was identified for contextual present factors. Fifteen studies investigated the effects of various contextual factors on child anticipatory distress during the painful medical procedure (e.g., type of admission and severity of procedure). Results vary based on the study.

Discussion

The purpose of this review was to summarize the findings of studies that examine the factors that predict anticipatory distress to painful medical procedures in children. The overarching goal of this review was to qualitatively synthesize the literature on the factors that predict anticipatory distress to painful medical procedures into a summary figure using predisposing, precipitating, perpetuating, and present factors as a framework. The following paragraphs will discuss key findings and patterns from the summary figure (Figure 2) of the review in the context of methodological differences and risk of bias within studies. Only factors with two or more included studies of a similar nature that can be found in the summary figure will be discussed. Finally, clinical implications, areas for future research based on the summary figure and limitations of the review will be highlighted.

Predisposing Factors

Child

There were some interesting patterns among the child predisposing factors. First, there is clear evidence that child psychopathology and difficult, fearful, or shy child temperament are individual child factors that increase the risk of child anticipatory distress. This finding is in line with developmental literature suggesting that children who have internalizing or externalizing problems have more difficulty regulating their affect (Bradley, 2003). Preexisting psychopathology or difficult temperament may be important factors to screen for prior to a medical procedure or surgery to have an understanding of how a child might respond or cope with the procedure. The risk of bias subsuming this factor was generally unclear because there were 6 low and 2 high-rated studies included.

Second, gender does not appear to play an important role in predicting anticipatory distress. Although some studies did find that girls experience higher anticipatory distress to medical procedures, the majority of studies did not find an effect. The studies that found an effect for girls had participants closer to pubertal age, which may have played a role.

Some predisposing child factors yielded inconclusive results. Despite the large body of research (43 studies) that examined the effect of age on child anticipatory distress, the research on this factor does not

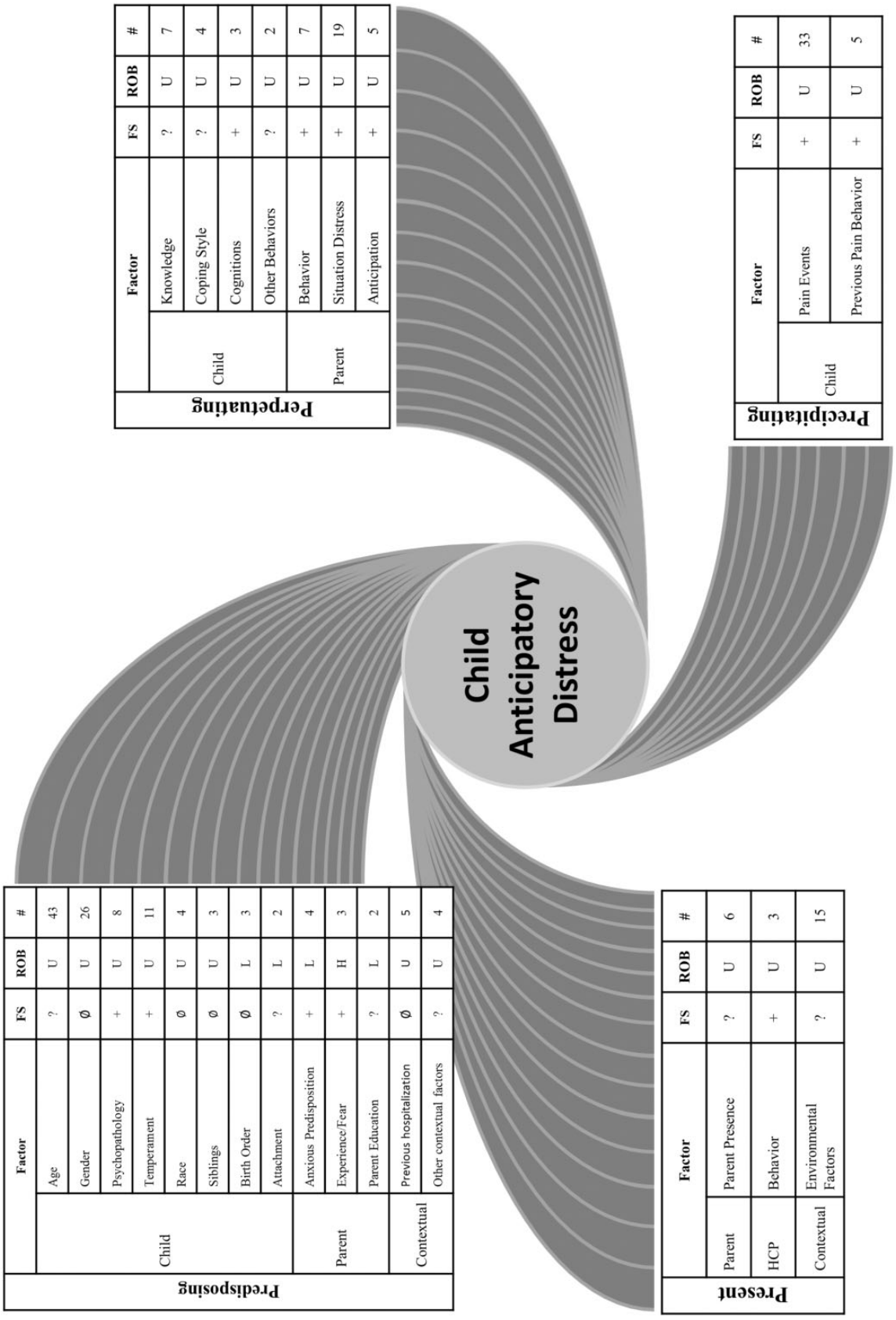

Figure 2. Summary figure of results. FS = findings synthesis; ROB = risk of bias; + = factor has a positive relationship with anticipatory distress; − = factor has a negative relationship with anticipatory distress; ∅ = no effect or significant relationship; ? = inconclusive results; U = unclear risk of bias; L = low risk of bias; H = high risk of bias; # = number of studies.

seem to converge. Almost half the studies showed no effect of age, while the other half suggests younger children experience higher anticipatory distress. The type of medical procedure did not seem to systematically differ between the two groups. Although the studies that did not find an effect of age were more likely to have a low risk of bias, it is difficult to make conclusions based on this. Methodological factors may also have contributed to differences in results as the majority of studies did not examine a discrete age range but rather averaged over large age ranges of up to 15 years. Examining a restricted age range may also have contributed to the lack of an effect. At this point, the results on age remain largely inconclusive, although the results from this review point toward younger children experiencing more anticipatory distress than older children. This is in line with the literature that indicates that younger children are more likely to be fearful and distressed and that this fear may increase and decrease over the course of childhood (American Psychiatric Association, 2013). It may also be the case that the relationship between age and anticipatory distress is nonlinear or co-varies with other factors. Future longitudinal or cross-sectional studies could provide some insight into whether age is an important factor in predicting child anticipatory distress.

Parent

Two parent predisposing factors that emerged as predicting increased child anticipatory distress are parent anxious predisposition and previous parent pain experience. The fact that a parent's own anxiety and fear/experiences with pain are related to the child's anticipatory distress directly supports the transmission of anxiety from parent to child. Previous work has hypothesized the mechanisms by which this occurs, such as through modeling and information transmission (Rachman, 1977; Vasey & Ollendick, 2000). It may be that parents are discussing or demonstrating their fear of pain as it relates to painful medical procedures, impacting the anticipatory distress of their children. Future experimental research could examine how transmission of fear of painful medical procedures occurs to develop targets for intervention. Risk of bias was variable across factors ranging the full gamut from low to unclear to high. Of note, the anxious predisposition has low risk of bias; thus, there is increased confidence in this finding.

Inconclusive results were found for the impact of parent education level on child anticipatory distress. The difference in finding may be due to the differences in education levels included in the studies. More research is needed in this area.

Context

Two contextual factors emerged under the predisposing domain. First, previous hospitalization was overall not found to have an effect on child anticipatory distress (only previous hospitalization of siblings did). It may be that hospitalization itself is not sufficient to lead to the development of fear but rather that negative experiences or vicarious fear are much more salient. The risk of bias for this factor was unclear as there was a mix of high and low rated studies. Few predisposing contextual factors have been examined and more research is needed.

Precipitating Factors

Child

Two broad factors were identified as factors that contributed to the onset of anticipatory distress to painful medical procedures. The first factor is general and specific negative pain events. Although the overall result is that previous negative pain events predict anticipatory distress, this was not uniformly the case across studies. Some reasons for this include risk of bias and sample size. The studies that found an effect of previous negative procedures largely had low risk of bias and large sample sizes. The studies that found a negative relationship between previous pain events and anticipatory distress were methodologically different in that they all involved short routine medical procedures such as insulin injections (Hanas et al., 2002; Howe, Ratcliffe, Tuttle, Dougherty, & Lipman, 2011) and immunotherapy injection (deVos et al., 2012). These types of procedures provide repeated exposure to the stimuli whereby eventually extinction of the fear occurs. Typically, developing children do not usually have daily exposure to needles or surgery to facilitate extinction, which may explain the difference in finding for the studies. Furthermore, previous work in child anxiety has demonstrated that direct conditioning is only one pathway to the development of anxiety problems in children (Vasey & Dadds, 2001). According to retrospective reports of adults with phobia, modeling and information transmission were the most common modes of fear acquisition with a minority reporting direct conditioning experiences (Vasey & Ollendick, 2000). This highlights that, although direct conditioning of a general or specific negative pain event may precipitate anticipatory distress for some children, multiple factors are at play, including the frequency and severity of the painful medical procedure.

The second factor that was identified as a child precipitating factor is previous child/adolescent behavior (five studies). There is evidence that a child or adolescent's previous behavior during a painful medical procedure will predict anticipatory distress at a future medical procedure. The risk of bias was rated as unclear due to one study with an unclear rating.

Perpetuating Factors

Child

Four child factors were identified as maintaining child anticipatory distress. First, child maladaptive cognitions were found to positively predict distress prior to a procedure with overall low risk of bias studies. Overall, children who perceived less control expected an aversive experience and appraised procedures as more threatening were more inclined to be distressed prior to those procedures. Given that child threat appraisal and perceived control predict child anticipatory distress, this highlights the importance of teaching children cognitive and behavioral coping strategies to manage their anticipatory distress. The overall risk of bias is unclear, reducing our confidence in these findings.

In terms of having more knowledge about the procedure, the overall results were inconclusive; however, three studies did show a decrease in anticipatory distress. The method, type of information, and developmental level of the knowledge provided may be important variables in whether the knowledge presented works. The overall risk of bias was unclear for this factor, reducing our confidence in the findings. There was inconclusive evidence for child coping style with unclear risk of bias, reducing confidence in these findings. There are specific child behaviors such as using nonprocedural talk, humor, and talking to a parent that were related to child coping before a procedure, while verbal resistance was found to be positively associated to child preoperative anxiety. The child behavior factor had an overall unclear risk of bias, reducing our confidence in these findings. More research on the child behaviors that are associated with coping before a painful medical procedure will help inform targets for intervention.

Parent

When examining the parent factors that perpetuate a child's anticipatory distress to painful medical procedures, an important pattern emerges. Across three factors examined, there was evidence that parent factors play a key role in maintaining the distress of children during painful medical procedures. Parent behavior during the procedure, parent situational distress/state anxiety, and parent anticipation of child distress had overall results predictive of child anticipatory distress. Although the findings of the studies were not completely uniform, the majority of studies highlighted the role that parents play in continuing child anticipatory distress. It has been argued that, particularly for infants and young children, the caregiver is the most important context in the pediatric pain setting (Pillai Riddell & Racine, 2009). Parental responding (modeling, overprotection, reinforcement, and encouragement) plays a key role in the development of anxiety (Vasey & Dadds, 2001). These results highlight the importance of engaging parents in interventions to help reduce child anticipatory distress. The risk of bias for the parent perpetuating factors was unclear, indicating reduced confidence in these findings.

Present Factors

Parent

A trend toward family-centered care has led to the increase of parental presence within pediatric healthcare settings. Although parent presence during child hospital stays have been associated with positive outcomes (Wright, Stewart, & Finley, 2010), parent presence during a painful medical procedure in this review had inconclusive results. However, two studies pointed to mechanisms that may underlie the effect of parental presence. Kain, Caldwell-Andrews, Maranets, Nelson, and Mayes (2006) found that the presence of a calm parent reduces preoperative anxiety, while the presence of an overly anxious parent does not. Given the transactional and individual factors that predict anticipatory distress, clinical recommendations for parental presence during a procedure should be based on characteristics of the parent and their ability to provide calm support rather than the blanket assumption that all parents should consistently be present or not present. More research should investigate the conditions under which parental presence is beneficial in reducing child anticipatory distress.

Health Professional

Our synthesis demonstrates that health care professional behavior does play an important role in predicting child anticipatory distress. Given the crucial role that healthcare professionals can play in the experience of children and families during painful medical procedures (Mahoney, Ayers, & Seddon, 2010), researchers should continue to examine distress reducing behaviors such as distraction that could be taught to health care professionals, as well as parents, as an intervention to reduce procedural distress prior to a painful medical procedure.

Contextual

Fifteen studies investigated the effects of various contextual factors on child anticipatory distress during the painful medical procedure. It is difficult to synthesize this research due to the varied contextual factors; however, some environmental factors (e.g., induction location and sensory stimulation) do seem to impact anticipatory distress. More research is needed to determine which contextual factors should be addressed in clinical practice.

Author's Conclusions and Clinical Implications

As outlined in developmental psychopathology theory (Cicchetti & Cohen, 1995), the development of anticipatory distress occurs through a dynamic interplay of factors, including individual child factors, parent factors, health professional factors, and their environment. There is no unique pathway that leads to the development of anticipatory distress but rather the interaction of predisposing, precipitating, perpetuating, and present factors over time leads to the onset and maintenance of distress.

In this review, we examined 31 factors that predict anticipatory distress. Children with preexisting anxiety and a difficult temperament were more likely to have anticipatory distress. Parents and children should examine patterns of past child behavior during painful procedures to better support and prepare children with these risk factors. Parent anxiety and parent's previous experiences with pain are also important predictors of anticipatory distress. This suggests that parents need to be aware of their own subjective experience of medical procedures and how to manage their own anxiety in medical contexts. Past pain events and previous child behavior are indicators of future anticipatory distress. Using adequate pain management is of utmost importance in reducing the likelihood of conditioning fear and anxiety. Child and parent emotional and cognitive factors serve to maintain or fuel anticipatory distress. These areas will be important targets for interventions. Finally, health professionals should be wary of engaging in distress promoting behavior such as verbal reassurance and criticism and are encouraged to use coping promoting behavior such as talking about things other than the procedure and engaging in distraction.

Limitations and Implications for Research

This review highlights important gaps where additional research is needed. As shown in Figure 2, factors that are depicted with a question mark have inconclusive evidence. All of these areas would benefit from additional research to investigate their impact on anticipatory distress. Additionally, there is a need for longitudinal and more complex methodologies to investigate the transactional nature of these factors. Future studies should also examine the interaction of multiple factors (i.e., temperament, previous pain experience, and parent behavior) to determine the relative contribution of these factors. Furthermore, many of the studies included in the review were found to have high risk of bias often through biased outcome assessors or poor quality measures used. Many of the factors (e.g., child level of anxiety, age, gender, parent anxiety, and previous pain experiences) cannot be randomized to participants to improve the quality of the methodology to test these factors. However, the knowledge of these proposed factors should be incorporated in randomized trials that test the efficacy of treatments of anticipatory distress. Having large age ranges in studies and not controlling for factors such as psychopathology (parent/child), previous pain experiences (parent/child) and parent soothing behaviors/coping strategies will continue to limit the value of randomized controlled trials because they do not attempt to accommodate the inherent variability of pain responses and the causes for the variability (Pillai Riddell et al., 2013).

Supplementary Data

Supplementary data can be found at: http://www.jpepsy.oxfordjournals.org/.

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