Critical Reviews

Prevalence and Predictors of Chronic Postsurgical Pain in Children: A Systematic Review and Meta-Analysis

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Abstract: Emerging research suggests that pain may persist longer-term for many children after major surgery, with significant effects on their health outcomes. This systematic review identified the prevalence of chronic postsurgical pain (CPSP) in children after surgery, and determined presurgical biomedical and psychosocial risk factors associated with CPSP prevalence or severity. Prospective studies assessing CPSP 3 to 12 months after surgery in children 6 to 18 years of age published in English in MedLine, EMBASE, PsycINFO, and Cochrane Database of Systematic Reviews since 1996 were eligible for inclusion. Of 16,084 abstracts yielded by the search, 123 full articles were assessed for eligibility, and 12 studies were included in the review. Overall quality of included studies assessed using the Quality in Prognostic Studies tool was low. On the basis of 4 studies with a total of 628 participants across all surgery types, median prevalence of CPSP across studies was 20% (25th percentile = 14.5%, 75th percentile = 38%) at 12 months after surgery. Presurgical pain intensity, child anxiety, child pain coping efficacy, and parental pain catastrophizing were the only presurgical factors identified as predictive of CPSP. Biological and medical factors assessed were not associated with CPSP in any study. Well designed studies examining prevalence and predictors of CPSP are critically needed in children.

Perspective: In this systematic review, the median prevalence of CPSP in children was 20% across studies. Presurgical pain intensity, and child and parent psychosocial factors predicted CPSP. Additional resources and interventions are needed for youth who report persistent pain after surgery.

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Key words: Postoperative pain, chronic pain, child, pediatric, persistent pain, operative.

Close to 5 million children undergo surgery in the United States each year, and many experience significant pain and distress during the initial days and weeks of recovery. Approximately one-half of children report moderate-severe pain in the hospital after surgery.5 However, relatively less is known about the short- and long-term recovery of children after surgery. Children often continue to report pain for months after surgery, and emerging research suggests that pain may persist longer-term for many children.

Chronic postsurgical pain (CPSP) is defined as pain lasting >3 months after surgery, that is not otherwise associated with preexisting problems or postoperative complications. CPSP is reported in 10 to 50% of adults undergoing major surgery.3,9 CPSP in adults is associated with higher functional disability, increased missed work days, and higher levels of depressive symptoms. Recent studies have investigated long-term outcomes in children undergoing surgery, showing that CPSP is associated with poorer health outcomes and with greater functional disability after surgery.22
Prevalence and Predictors of Pediatric CPSP

Types of Studies

We only considered studies that were published, peer-reviewed reports, written in English. Non-English studies were excluded because we did not have resources to interpret foreign language articles. We considered different study designs including cross-sectional, case series, case-control, and cohort studies that included more than 10 participants. Single case reports, retrospective studies (e.g., chart review), and intervention studies were excluded. Only studies that reported pain between 3 and 12 months after surgery were eligible for inclusion.

Types of Outcomes

Prevalence of CPSP Pain

The primary outcome was the presence of pain 3 to 12 months after surgery. We did not define the cutoff for pain presence but rather abstracted the definition used in each individual study. We abstracted presence of pain as well as severity of pain. If multiple reports of pain were assessed in this period, each was extracted. If self-report was not available, we extracted parent report or nurse report of child pain.

Risk Factors for CPSP Pain

Biological (age and sex), medical (baseline pain severity and location), and psychosocial (presurgical child anxiety, child pain catastrophizing, child depression, child sleep patterns, parent anxiety, and parent pain catastrophizing) factors were extracted. Only risk factors that were assessed in the presurgery period were extracted and included in the analyses.

Search Strategy

Cochrane Database of Systematic Reviews, MedLine, EMBASE, and PsycINFO were searched for peer-reviewed studies published from January 1996 to June 2016. This cutoff was chosen on the basis of major advances in surgical and anesthetic techniques occurring in the early 1990s making historic reports less relevant to current practice. A MedLine search strategy was developed first and was adapted for other search engines (see Supplementary Appendix 1). Our search strategy includes terms for children/adolescents, surgery, and pain. The reference list of each included study was manually searched for additional reports potentially meeting inclusion criteria. We also conducted a citation search for each included study to identify other potential studies for inclusion.

Data Collection and Analysis

Study Selection

One reviewer screened the abstracts to identify potential studies. A second reviewer screened 10% of all abstracts. There was a high level of agreement (99.4%) on screening. Two reviewers then assessed the full articles of potentially eligible studies for inclusion in the systematic review. Disagreements were discussed and resolved with a third author.

Methods

Inclusion Criteria

Types of Participants

We included studies of children 6 to 18 years undergoing surgery. Children had to receive general or regional anesthesia at a hospital or surgery center. Diagnostic and noninvasive procedures were excluded. Studies investigating children undergoing cancer surgery (malignant) or with a neurological disability were excluded because of multiple confounding factors in these populations. Age 6 years was chosen as the lower bound as the age at which children reliably self-report pain. Studies extending beyond the eligible age limits were considered for inclusion if most participants were within the eligible age range, or if data were reported separately for children in the eligible age range.

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Data Extraction

One reviewer extracted data from studies meeting the inclusion criteria, which was checked by a second reviewer. We extracted study characteristics (study type, sample size, surgery type), participant characteristics (age, sex, surgery type), the presence of pain 3 to 12 months after surgery, pain severity between 3 and 12 months, and presurgery biopsychosocial risk factors.

Risk of Bias

Risk of bias of included studies was assessed by 2 reviewers using the Quality in Prognosis Studies tool. Disagreements between the 2 reviewers were discussed and a third reviewer arbitrated if agreement was not met. Each study was assessed on 6 items; study participation, study attrition, prognostic factor measurement, confounding measurement and account, outcome measurement, and analysis and reporting. Risk of bias assessments followed guidelines by Hayden and colleagues. First, for study participation, we allocated a low risk of bias if there was a description of the time frame that the study recruited participants, the inclusion/exclusion criteria, the source populations, and the baseline sample. We assessed moderate risk of bias if the study partially met the requirements for low risk of bias. We allocated a high risk of bias if studies did not report the details mentioned previously, or if there was a low participation rate, a reported sex or age distribution that is unusual for the population being investigated, or if there was biased recruitment of the sample. We judged a study to have high risk of bias if the sample was poorly described.

We assessed low risk of bias for study attrition if authors reported participant attrition throughout the study and reasons for attrition, and included a completer versus noncompleter analysis to determine whether there were any differences. High risk of bias was given to studies not meeting this criterion.

Low risk of bias for prognostic factors was given to studies that used validated measures to investigate prognostic factors (eg, anxiety, coping efficacy) to predict pain at follow-up. Studies that did not aim or assess prognostic factors were not evaluated on this outcome.

We judged low risk of bias for outcome measurement if a valid and reliable pain measure was used, if the authors described when the pain measurement occurred, and if outcomes were assessed consistently across participants. Measures commonly used within the population (eg, visual analog scale, NRS) with established psychometric properties were considered valid and reliable. Alternatively, if authors referenced an article supporting the psychometrics of the measure within a similar population, we considered these measures as valid and reliable. Moderate or high risk of bias was allocated to studies that did not describe these details.

Low risk of bias was allocated to studies that did include confounding factors (eg, age and sex) in their analyses. We judged confounding factors as high risk of bias if none were controlled for in the analyses.

Finally, low risk of bias was allocated to studies reporting appropriate statistical tests for their research question. High risk of bias was given to studies that only used univariate analyses to investigate relationships between variables, or if data analyses were not appropriate for the variables assessed.

Data Synthesis

Prevalence of CPSP

We aimed to describe the median, 25th percentile, and 75th percentile of prevalence of postsurgical pain between 3 and 12 months across studies. Only a subgroup of studies reported prevalence of pain after surgery. However, all studies within this subgroup reported prevalence of CPSP at the 12-month follow-up. Therefore, we chose to summarize prevalence for this time point. Because of limited reporting of pain prevalence in the included studies, we also summarized reported pain severity at follow-up. Where overlapping samples were presented in different studies, we only described the first published study, but extracted from other secondary studies if additional data were available that were not included in the primary study. For studies extracting data from registries, we highlighted where overlapping or inflated estimates may have occurred.

Subgroup analyses. Where enough data were available (at least 2 studies), we aimed to pool prevalence of CPSP according to individual surgery type. In particular, we identified the following surgery types for subgroup analysis: spinal fusion, thoracotomy, laparotomy, chest wall surgery, inguinal hernia repair, amputation, and sternotomy. However, data were only available for mixed surgery samples and spinal fusion surgery. No studies meeting our inclusion criteria were available for other surgery types.

Risk Factors for CPSP

Because of the lack of data, we were unable to perform meta-analysis on biopsychosocial risk factors. Therefore, we qualitatively summarized baseline biological, medical, and psychological risk factors to predict the presence or severity of CPSP (3–12 months). We were primarily interested in sex, age, presurgical pain severity, and presurgical child and parent anxiety, depression, and pain catastrophizing.

Results

Search Results

Our search criteria were broad, producing a large number of abstracts for initial sifting. The search produced 16,084 abstracts, of which 584 were duplicates (Fig 1). We identified and read 123 full articles for inclusion, and included 13 in our final sample. Many of the studies excluded in the first sift of the abstracts were retrospective (eg, medical chart review), examined nonsurgical procedures (dental extraction), included adults, or did not assess pain within our defined time frame (eg, only assessed in-hospital pain). Of the 13 articles that met our inclusion criteria, 2 reported on
the same study.\textsuperscript{17,18} Therefore, 12 studies are included in our review.\textsuperscript{2,4,11-15,17,18,20,22-24}

**Study Characteristics**

The 12 studies included in our systematic review were published between 2010 and 2016.\textsuperscript{2,4,11-15,17,18,20,22-24} Study characteristics are presented in Table 1. There were 1,835 children and adolescents included in these studies, however, there may be some duplication because of several of the studies using data from central registries. Of the 10 studies that reported a mean age, children were an average of 14.37 years old at the time of surgery. Seven studies exclusively focused on patients

<table>
<thead>
<tr>
<th>Study</th>
<th>N</th>
<th>Mean Age (Range), Y</th>
<th>Sex: Female/Male</th>
<th>Surgery Type</th>
</tr>
</thead>
<tbody>
<tr>
<td>Carreon et al\textsuperscript{2}</td>
<td>887</td>
<td>14.3 (10–18)</td>
<td>735/152</td>
<td>Spinal fusion</td>
</tr>
<tr>
<td>Connelly et al\textsuperscript{4}</td>
<td>50</td>
<td>14.5 (11–17)</td>
<td>41/9</td>
<td>Posterior spinal fusion</td>
</tr>
<tr>
<td>Landman et al\textsuperscript{11}</td>
<td>295</td>
<td>NR (8–22)*</td>
<td>NR*</td>
<td>Spinal fusion</td>
</tr>
<tr>
<td>Lillehei et al\textsuperscript{12}</td>
<td>44</td>
<td>14.7 (10–18.8)</td>
<td>28/16</td>
<td>Ilioanal pull through</td>
</tr>
<tr>
<td>Lind et al\textsuperscript{13}</td>
<td>20</td>
<td>12.5 (8–16)</td>
<td>11/9</td>
<td>Medial patellofemoral ligament reconstruction (knee)</td>
</tr>
<tr>
<td>Mariconda et al\textsuperscript{14}</td>
<td>87</td>
<td>14.8 (11–22)</td>
<td>77/10</td>
<td>Spinal fusion</td>
</tr>
<tr>
<td>Nishimura et al\textsuperscript{15}</td>
<td>12</td>
<td>14.4 (12–17)</td>
<td>0/12</td>
<td>Osteochondral transplant (donor knee evaluated)</td>
</tr>
<tr>
<td>Page et al\textsuperscript{17,18}</td>
<td>83</td>
<td>13.8 (8–18)</td>
<td>56/27</td>
<td>Major orthopedic and general surgery</td>
</tr>
<tr>
<td>Pellegrino et al\textsuperscript{19}</td>
<td>33</td>
<td>15.6 (11–20)</td>
<td>3/12</td>
<td>Posterior spinal fusion</td>
</tr>
<tr>
<td>Rabbits et al\textsuperscript{22}</td>
<td>60</td>
<td>14.7 (10–18)</td>
<td>40/20</td>
<td>Major orthopedic and general surgery</td>
</tr>
<tr>
<td>Sieberg et al\textsuperscript{23}</td>
<td>190</td>
<td>14.4 (8–21)</td>
<td>137/53</td>
<td>Spinal fusion</td>
</tr>
<tr>
<td>Stromqvist et al\textsuperscript{24}</td>
<td>74</td>
<td>17 (12–17)\textsuperscript{f}</td>
<td>43/31</td>
<td>Surgery for lumbar disk herniation</td>
</tr>
</tbody>
</table>

*Median age reported.
*Demographic characteristics only provided for full sample of 1,433 patients and not for 295 included in the analyses.

**Table 1. Characteristics of Included Studies**
undergoing spine surgery, of which 6 included patients with scoliosis and 1 included patients with lumbar disk herniation. In addition, 2 further studies examined youth undergoing a mix of different surgeries, however, the most participants underwent spine surgery. Of the remaining 3 studies, 1 examined outcomes after ilioanal pull through (a major abdominal procedure), and 2 small studies examined outcomes after 2 specific knee surgeries. All of these procedures are major inpatient surgeries with the exception of the knee procedures, which are usually performed as outpatient procedures. Most patients in the included studies were female, which is consistent with the sex differences in rates of scoliosis.

Eight studies were prospective cohort studies and 4 studies analyzed data from prospectively collected patient registries. Three of these database studies used a registry for scoliosis surgery from the Spinal Deformity Study Group, and 1 used data on patients having lumbar spine surgery from the Swedish National Spine Register. Sieberg et al included 190 patients from Boston Children’s Hospital between 2003 and 2007. Landman et al included 295 patients enrolled in the database from the United States since 2003 (end date not provided). Carreon et al included 887 patients from sites within the United States and Europe, however, date ranges were not provided. There is likely to be significant overlap in data among these 3 studies.

**Risk of Bias**

Of the 12 studies included in the review, 4 were judged as having low risk of bias for study participation because they provided full descriptions of their study participants. Five were assessed as moderate risk of bias, and 3 were judged to have high risk of bias providing an insufficient account of recruitment and sample (see Fig 2 and Supplementary Appendix 2 for risk of bias assessments). Regarding study attrition, only 1 study provided an adequate description of attrition and included a completer versus noncompleter analysis. Five studies provided partial description of attrition, and were judged to have moderate risk of bias, and the remaining 6 studies did not provide descriptions of attrition and were rated as high risk of bias.

Six studies included prognostic factors, of which 4 were judged to be low risk of bias. We judged 2 studies to have a moderate risk of bias for this category, because either some measures were not valid or reliable, or a domain of an outcome measure was used for the prognostic factors and a different domain of the same measure was used to assess the outcome. Fourth, we assessed risk of bias for outcome measurement. We judged 3 studies to use valid and reliable measures of pain assessment, applied consistently across participants. Most studies did not describe when or how measurements were performed and were judged to have moderate risk of bias (n = 8). We judged 1 study to have high risk of bias because the authors did not describe how pain was assessed or the pain measure used.

### Table 1. Quality in Prognosis Studies Risk of Bias for Included Studies

<table>
<thead>
<tr>
<th>Study</th>
<th>Study participation</th>
<th>Prognostic factors</th>
<th>Outcome measurement</th>
<th>Confounding factors</th>
<th>Statistics and reporting</th>
</tr>
</thead>
<tbody>
<tr>
<td>Carreon et al. 2</td>
<td>Low risk of bias</td>
<td>Moderate risk of bias</td>
<td>Low risk of bias</td>
<td>Low risk of bias</td>
<td>N/A</td>
</tr>
<tr>
<td>Connelly et al. 4</td>
<td>Moderate risk of bias</td>
<td>Moderate risk of bias</td>
<td>Low risk of bias</td>
<td>Low risk of bias</td>
<td>N/A</td>
</tr>
<tr>
<td>Landman et al. 11</td>
<td>Low risk of bias</td>
<td>Low risk of bias</td>
<td>Low risk of bias</td>
<td>Low risk of bias</td>
<td>N/A</td>
</tr>
<tr>
<td>Lillehei et al. 12</td>
<td>Low risk of bias</td>
<td>Low risk of bias</td>
<td>Low risk of bias</td>
<td>Low risk of bias</td>
<td>N/A</td>
</tr>
<tr>
<td>Lind et al. 13</td>
<td>Low risk of bias</td>
<td>Moderate risk of bias</td>
<td>Low risk of bias</td>
<td>Low risk of bias</td>
<td>N/A</td>
</tr>
<tr>
<td>Mariconga et al. 14</td>
<td>Low risk of bias</td>
<td>Moderate risk of bias</td>
<td>Low risk of bias</td>
<td>Low risk of bias</td>
<td>N/A</td>
</tr>
<tr>
<td>Nishimura et al. 15</td>
<td>Low risk of bias</td>
<td>Moderate risk of bias</td>
<td>Low risk of bias</td>
<td>Low risk of bias</td>
<td>N/A</td>
</tr>
<tr>
<td>Page et al. 17, 18</td>
<td>Low risk of bias</td>
<td>Moderate risk of bias</td>
<td>Low risk of bias</td>
<td>Low risk of bias</td>
<td>N/A</td>
</tr>
<tr>
<td>Pellegrino et al. 20</td>
<td>Low risk of bias</td>
<td>Moderate risk of bias</td>
<td>Low risk of bias</td>
<td>Low risk of bias</td>
<td>N/A</td>
</tr>
<tr>
<td>Rabbitts et al. 22</td>
<td>Low risk of bias</td>
<td>Moderate risk of bias</td>
<td>Low risk of bias</td>
<td>Low risk of bias</td>
<td>N/A</td>
</tr>
<tr>
<td>Sieberg et al. 23</td>
<td>Low risk of bias</td>
<td>Moderate risk of bias</td>
<td>Low risk of bias</td>
<td>Low risk of bias</td>
<td>N/A</td>
</tr>
<tr>
<td>Stromqvist et al. 24</td>
<td>Low risk of bias</td>
<td>Moderate risk of bias</td>
<td>Low risk of bias</td>
<td>Low risk of bias</td>
<td>N/A</td>
</tr>
</tbody>
</table>

**Figure 2.** Quality in Prognosis Studies risk of bias for included studies. Abbreviation: N/A, not applicable.

Most studies did not include confounding demographic factors in their analyses and were therefore judged to have a high risk of bias (n = 7). Three studies did include confounding factors, which we judged to have low risk of bias and 2 studies had moderate risk of bias. Finally, 3 studies used appropriate statistical methods, which were reported and were judged as low risk of bias. Three studies used partially appropriate statistical methods, which we judged to have moderate risk of bias. Six studies only used univariate analyses, which were not adequate to answer the research questions proposed and so were allocated as high risk of bias.

### Prevalence of Chronic Postsurgical Pain

Four studies with a total of 628 participants across all surgery types reported prevalence of CPSP. The median prevalence of CPSP across studies was 20% (25th percentile = 14.5%, 75th percentile = 38%) at 12 months after surgery. Prevalence ranged widely among studies, likely because of inconsistent definitions of CPSP. Higher prevalence estimates (54%) were described for presence of any pain, whereas lower prevalence estimates (11–22%) were reported for moderate-severe pain. Studies with higher risk of bias reported prevalence estimates (11% and 54%) which diverged most from the median prevalence across studies, whereas studies...
with low to moderate risk of bias reported prevalence estimates (18% and 22%) close to the median prevalence.

**Spinal Fusion**

Two studies reported rates of pain of 11%\textsuperscript{23} and 54%\textsuperscript{11} 12 months after spinal fusion. Both of these studies had moderate to high risk of bias in most categories, and draw from the same database and therefore we did not calculate summary statistics for pain prevalence for this surgery type.

**Mixed Major Surgeries**

On the basis of 2 studies including mixed major surgeries, the median prevalence of CPSP after mixed major surgeries was 20%. These 2 prospective studies reported rates of pain of 18%\textsuperscript{22} and 22%\textsuperscript{18} at 1 year after major general and orthopedic surgeries. Both studies reported similar rates of pain at earlier assessments (3 and 4 months). These studies both had low to moderate risk of bias.

**Chronic Postsurgical Pain Severity**

Nine studies provided summary statistics for pain severity 3 to 12 months after surgery, presented in Table 2. Measures used for assessing pain varied across studies. Across the 3 studies using a 0 to 10 numeric rating scale (NRS), low median pain intensity was found at 12 months (median = 1.5 of 10). Across the 3 studies using the pain domain from the Scoliosis Research Society (SRS) measure, favorable pain scores were reported on the pain domain at 12 months (median = 4.3 of 5, range 1–5, higher score indicates better pain outcome).

**Measurement of Postsurgical Pain Severity**

Of the 12 studies included in the review, 5 used standard measures of pain intensity (NRS, and visual analog scale [VAS]). The remaining 7 used quality of life measures (SRS-22, SRS-23, SRS-30, and Short Form-36) to assess pain. Five of these 7 studies used validated scores from the pain domains of the quality of life measures, and 2 used a single pain item from the SRS measures assessing pain frequency. Most of the studies did not indicate the time frame assessed (ie, current, past week, past month), or whether worst, least, usual, or provoked pain were reported.

**Risk Factors for Chronic Postsurgical Pain**

Three studies examined presurgical risk factors for CPSP, presented in Table 3. Two studies examined associations between biological, medical, and psychological factors and presence or severity of postsurgical pain 3 to 12 months after surgery.\textsuperscript{4,22} One additional study examined age and sex as predictors of presence of chronic pain at 6 and 12 months after surgery.\textsuperscript{18}

### Table 2. Chronic Pain Outcomes 3 to 12 Months After Surgery

<table>
<thead>
<tr>
<th>Study</th>
<th>Pain Measure*</th>
<th>Time Point (Mo)</th>
<th>Mean Pain Severity (SD, Range)</th>
<th>Chronic Pain Definition</th>
<th>Pain Prevalence</th>
</tr>
</thead>
<tbody>
<tr>
<td>Carreon et al\textsuperscript{2}</td>
<td>SRS-22 pain domain</td>
<td>12</td>
<td>4.35 (.61)</td>
<td>NR</td>
<td>NR</td>
</tr>
<tr>
<td>Connelly et al\textsuperscript{4}</td>
<td>VAS 0 to 100, typical pain in past month</td>
<td>3</td>
<td>13.91 (18.85, 0–70)</td>
<td>NR</td>
<td>NR</td>
</tr>
<tr>
<td>Landman et al\textsuperscript{11}</td>
<td>SRS-22 pain item, pain in past month</td>
<td>6</td>
<td>11.57 (17.56, 0–82)</td>
<td>NR</td>
<td>Mild-severe (rarely to very often) 158 of 295 (53.6%)</td>
</tr>
<tr>
<td>Lillehei et al\textsuperscript{2}</td>
<td>SF-36 bodily pain domain</td>
<td>12</td>
<td>87.2 (13.8)</td>
<td>NR</td>
<td>NR</td>
</tr>
<tr>
<td>Lind et al\textsuperscript{13}</td>
<td>NRS 0 to 10, pain when walking</td>
<td>12</td>
<td>1.5 (1.3)</td>
<td>NR</td>
<td>NR</td>
</tr>
<tr>
<td>Mariconda et al\textsuperscript{14}</td>
<td>SRS-23 pain domain</td>
<td>12</td>
<td>3.8 (.6)</td>
<td>NR</td>
<td>NR</td>
</tr>
<tr>
<td>Nishimura et al\textsuperscript{15}</td>
<td>VAS 0 to 10, knee pain</td>
<td>6</td>
<td>0 (0, 0–0)</td>
<td>NR</td>
<td>NR</td>
</tr>
<tr>
<td>Page et al\textsuperscript{17,18}</td>
<td>NRS 0 to 10, current pain intensity</td>
<td>6</td>
<td>1.9 (1.3)</td>
<td>Moderate-severe (≥4 of 10) 14 of 61 (23.1%)</td>
<td>13 of 59 (22.0%)</td>
</tr>
<tr>
<td>Pellegrino et al\textsuperscript{20}</td>
<td>SRS-30 pain domain</td>
<td>3</td>
<td>3.18 (.14)</td>
<td>NR</td>
<td>NR</td>
</tr>
<tr>
<td>Rabbitts et al\textsuperscript{22}</td>
<td>NRS 0 to 10, daily pain assessment for 7 d</td>
<td>4</td>
<td>1.2 (1.7)</td>
<td>Late pain recovery trajectory\textsuperscript{\textdagger}</td>
<td>11 of 60 (18.3%)</td>
</tr>
<tr>
<td>Sieberg et al\textsuperscript{23}</td>
<td>SRS-30 pain item, pain in past month</td>
<td>12</td>
<td>1.1 (1.3)</td>
<td>Moderate-severe\textsuperscript{\textdagger}</td>
<td>19 of 169 (11%)</td>
</tr>
<tr>
<td>Stromqvist et al\textsuperscript{24}</td>
<td>SF-36 bodily pain domain</td>
<td>12</td>
<td>NR</td>
<td>NR</td>
<td>NR</td>
</tr>
</tbody>
</table>

Abbreviations: NR, not reported; VAS, visual analog scale; SF-36, Short Form-36.

NOTE. SRS uses a 1 to 5 scale with higher scores on pain domain indicating better pain-related quality of life; higher scores of pain items indicate lower pain frequency. Higher scores on a VAS and NRS indicate higher pain intensity. SF-36 uses a 0 to 100 scale, with higher scores indicating higher functioning.

*Time frame of pain ratings provided if reported.

\textdagger}Trajectory on the basis of statistical modeling of pain over 12 months.

\textdagger}Cutoff/range NR.
Biological Factors

Three studies examined age and sex as predictors of CPSP in children.4,18,22 Contrary to hypotheses, significant relationships were not found.

Medical Factors

Two studies examined associations between presurgery pain intensity and CPSP, with conflicting results. Connelly et al4 reported significant associations between presurgery pain and postoperative pain whereas Rabbitts et al22 did not. Connelly et al4 also examined other clinical factors including presurgery body mass index, scoliosis severity (Cobb angle), and time since scoliosis diagnosis, reporting that none of these factors were associated with postsurgical pain outcomes.

Psychosocial Factors

Two studies examined relationships between presurgery psychosocial factors and chronic postoperative pain3,22 and reported significant relationships. Connelly et al4 reported greater presurgery child anxiety and poorer pain coping efficacy, but not mood or family income, were associated with slower rate of decline in pain over 6 months after surgery. Rabbitts et al22 examined child as well as parental factors, reporting that presurgery parental pain catastrophizing was associated with CPSP. However, child pain catastrophizing was not associated with chronic postoperative pain.

Discussion

Our systematic review findings provide prevalence estimates of CPSP in children 3 to 12 months after surgery. The estimated median prevalence of CPSP was 20% across included studies, which is similar to estimated rates in adults. Only 2 of the 12 studies included in this review assessed biopsychosocial risk factors of CPSP, before surgery. Presurgical pain intensity and child and parent psychosocial factors were the only presurgical factors identified as predictive of CPSP. Biological factors (age and sex) and other medical factors assessed were not associated with CPSP in any study. To our knowledge, this is the first systematic review of prevalence and risk factors of CPSP in children.

Despite a broad search strategy, we identified few studies meeting inclusion criteria. Among the 12 included studies, only 4 studies reported the prevalence of CPSP and only 3 assessed presurgery factors as predictors of chronic pain after surgery. Baseline risk factors for CPSP were inconsistent across studies, partly because of the limited number of studies and the small sample sizes. Equivocal findings of the association between presurgery pain and CPSP in 2 studies may be related to differences in how pain was analyzed in the trajectory studies. Connelly et al4 examined slope of pain scores after surgery, whereas Rabbitts et al22 examined trajectory groups on the basis of pattern over time. Similarly, in the only 2 studies examining presurgery psychosocial risk factors multiple differences in study design, risk factors, and study outcomes may account for differing findings. Connelly et al4 examined only child psychosocial factors, identifying child anxiety and pain coping efficacy as predictors, whereas Rabbitts et al22 examined parent as well as child factors with parent catastrophizing as the only significant predictor. Whereas Page et al17,18 assessed psychosocial factors after surgery, findings were consistent with Rabbitts et al22 in that parent factors and not child factors were associated with presence of CPSP. The literature is currently too small to draw any consistent conclusions as to which risk factors are most likely to be important in the transition to CPSP, however initial findings indicate psychosocial factors warrant further study. Larger sample sizes are needed to comprehensively assess presurgical risk factors.
factors, and to understand the magnitude of risk associated with these factors.

Although extensive research has examined CPSP in adults, the paucity of literature in children highlights the critical need for research examining longer-term pain outcomes in children after surgery. Although our search strategy began in 1996, no studies before 2010 met our inclusion criteria, reflecting that pediatric postsurgical pain has received increasing attention in the past 6 years. Most studies did not describe CPSP prevalence, but rather presented mean pain severity at 3-to-12-month follow-up time points. However, by averaging across all participants in a sample, important data on individual pain patterns are lost. For example, studies that reported mean pain severity as well as prevalence of CPSP reported low levels of pain across the whole sample (mean pain intensity 1.1 ± 1.3 and 1.6 ± 1.1 on a 0–10 NRS). However, they identified a significant portion of individuals experiencing CPSP at 12 months (18% and 22%, respectively). Identifying individual patterns of clinically meaningful pain at follow-up periods may enhance understanding of pain patterns in children after surgery.

Most studies assessing CPSP included patients undergoing spinal fusion. As a group, pediatric spine surgery has taken the lead in this area, by including pain as one of the domains in the widely used health-related quality of life outcome measure (SRS measures), and by establishing international registries for collection of long-term postsurgical outcomes data. This work has contributed to recognition of high rates of pain and the need for research aimed at improving pain outcomes in youth undergoing spine surgery. However, the interpretation of these database studies is problematic because of a lack of clarity regarding overlapping samples between studies.

A significant barrier to summarizing research in this area is the variation in the definition used for CPSP. A wide variety of measures and cutoffs were used to assess chronic pain after surgery. Similar issues have been raised in the literature assessing adult postsurgical pain, particularly the varied duration after surgery considered as chronic. Partly this is because of the varying duration of what can be considered normal healing after surgery. Furthermore, common definitions used in adult CPSP research include exclusion of a preexisting condition or complications. However, it can be challenging to differentiate this in surgical populations. Moreover, definitions of CPSP currently do not include pain frequency, pain intensity, or interference due to pain, which are important when identifying clinically meaningful pain. The current definition of CPSP is persistent pain of >3 months’ duration after surgery. However, definitions incorporating effect on the patient’s physical, psychological, or socioeconomic well-being have been proposed.

Notwithstanding these challenges, a unifying definition of CPSP across research and clinical practice is essential, which will require consistent criteria that may be derived from epidemiological studies of chronic pain prevalence using multidimensional pain and pain effect measures. Further, therapeutic clinical trials would also be facilitated by use of standard inclusion/exclusion criteria for individuals with CPSP.

A goal of our systematic review was to provide an assessment of study quality and we found that the overall quality of studies of CPSP was low. In particular, many studies did not take into account confounding variables (eg, demographic characteristics) in their analyses, meaning we do not know whether findings could be due to other variables. The use of univariate analyses were used frequently within the included studies despite aiming to answer research questions that required more sophisticated testing. On a broader level, many of the included studies did not report transparent study details (eg, sample description, flow diagrams of participant flow, comprehensive analysis plans, power calculations, full statistics) that would allow for assessing the rigor of the methodology. Future studies in this area will be enhanced by use of more rigorous study designs with validated outcome assessments, which will increase transparency, allow for synthesizing data across studies, and increase interpretability of study findings. Despite the overall low quality of studies, there were 3 studies that scored low risk of bias on at least half of the items.

**Limitations of This Review**

There are some limitations of our review that should be considered when interpreting the findings. First, we did not extract studies that included broad quality of life measures without explicitly referring to assessment of ‘pain’ in the abstract. It is possible that health-related quality of life measures that assess bodily pain were not included in our review. Second, an overall small number of studies met inclusion criteria. Many studies identified in our search did not meet our inclusion criteria regarding study design, participants, and pain assessment at 3 to 12 months. There are additional studies that included mixed samples of children or older adolescents as well as adults but unfortunately child outcomes could not be extracted separately. Third, some of our planned analyses could not be conducted because of the heterogeneity of pain measures used and limited available data reported. For example, we were not able to examine differences according to surgery type, which may have contributed to the variability in results.

**Implication for Practice and Research**

Our review highlights several areas that can be addressed through future research and practice. First, reporting standards of postsurgical studies should be improved. Studies should report on basic sample characteristics, refusal rate, attrition, and provide a clear description of the data analysis plan. Confounding variables should be considered, and statistics should be reported according to established guidelines (eg, Strengthening the Reporting of Observational Studies in Epidemiology). This will increase transparency and quality of research conducted. At a minimum, age and sex should be included in analyses assessing postsurgical pain in youth. Second, standardized and developmentally
appropriate pain measures should be agreed upon and
used across practice and research. The SRS question-
naires were used in some studies included in this report,
which is useful for children undergoing scoliosis surgery. However, the pain domain includes questions that over-
lap (eg, pain in the past month, pain in the past 6
months, and pain at rest), and does not provide an
assessment of pain intensity. In other studies, varying
pain intensity VASs using different anchors were used.
This heterogeneity across studies means that meta-
analyses are difficult to perform and produce wider con-
fidence intervals. We recommend using the VAS (0–100)
or a NRS of 0 to 10, which are both reliable and valid in
children older than 6 years of age, with anchors
‘worst pain possible’ and ‘no pain.’ In addition,
postsurgical studies that include adults as well as
children should report outcomes separately according
to age group.

With regard to practice, our review shows that pain
lasting for longer than 3 months occurs in a sizeable pro-
portion of youth undergoing major surgery and there
are identifiable baseline risk factors that may predict
child postsurgical pain. However, despite these data
showing the persistence of postsurgical pain, there are
currently few resources available for providing pain
management and support to children and families after
surgery. Further research is needed to replicate findings
of the risk factors identified in this study, and testing of
additional risk factors. Additional resources and inter-
ventions should be developed and implemented for
these youth who report persistent pain after surgery.
Further, although our review did not assess pain re-
ported after 12 months postsurgery, it is likely that these
children report pain beyond this marker and potentially
into adulthood.

Conclusions
In summary, this systematic review identified similar
rates of CPSP in children as have been reported in adults.
However, the findings are limited by the paucity and
poor quality of studies. Well designed studies examining
prevalence and predictors of chronic pain after surgery
are critically needed in children.

Acknowledgments
The authors thank Susan Klawansky, MLS, AHIP of Se-
attle Children’s Library and Information Commons for
consultation on search design, and assistance conducting
the search.

Supplementary Data
Supplementary data related to this article can be
found online at http://dx.doi.org/10.1016/j.jpain.2017.
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