







Lumbar Epidural versus Caudal Epidural for Postoperative Analgesia After Lower Extremity Osteotomy Surgery in Pediatric Patients with Osteogenesis Imperfecta: A Propensity-Matched Cohort Analysis in a Single-Center Over 9 Years

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Purpose: Although pediatric epidural analgesia is a well-established technique used perioperatively. It is unclear whether a lumbar or caudal epidural is suitable for osteogenesis imperfecta (OI) patients, which may be associated with brittle bones and spine deformity. We conducted a retrospective study to investigate and compare the efficacy of the two continuous epidural techniques in pediatric patients undergoing lower extremity osteotomy surgery using a propensity score-matched analysis (PSMA).

Patients and Methods: A total of 274 patients were included. Patients' age, weight, and height were adjusted using PSMA. 90 patients were matched for further analysis, with 45 patients in the lumbar epidural group (Group L) and 45 patients in the caudal epidural group (Group C). Pain scores were categorized into three grades: mild (0–3), moderate (4–6), and severe (7–10), and compared between the two groups. Additionally, operation time, operation site, blood loss, scoliosis, oral analgesic medications, and catheter or nerve-related complications were compared.

Results: There were no significant differences in operation time, operation site, scoliosis, and blood loss between the two groups. The percentage of moderate to severe pain during movement was significantly higher in Group L than in Group C, with 37.5% versus 17.5% on the second-day post-operation ($P=0.039$). However, no statistically significant difference was observed on other days. Additionally, there was no significant difference in oral medication consumption and complications between the two groups.

Conclusion: Both lumbar and caudal epidural analgesia can be effectively used postoperatively, and a caudal epidural should be considered where performing a lumbar epidural is challenging in OI pediatric patients.

Plain Language Summary: Osteogenesis imperfecta (OI) is a rare genetic disorder that affects the body's connective tissues, particularly the bones and ligaments. It is caused by abnormalities in type I collagen, which leads to skeletal fragility known as "brittle bones". This fragility can cause various issues, including an increased risk of fractures from minor trauma, limb deformities, and unusual fractures such as vertebral compressions. OI patients may also experience spinal manifestations such as scoliosis and kyphosis.

Lumbar epidural analgesia has been found to be effective in providing pain relief for surgeries that involve the lower extremities. Additionally, caudal epidural analgesia has also demonstrated its effectiveness in providing postoperative analgesia for surgeries that affect the lower limbs. However, there is still debate about the safety of epidural analgesia in patients with skeletal dysplasias, especially those with OI. Despite this uncertainty, our center, which was supported by the Rare Diseases Public Welfare Organization, has successfully used epidural analgesia since 2015 in the southern part of China for OI surgeries.

We conducted a retrospective study to share our experiences of nine years of practice and compare lumbar epidural with caudal epidural using a propensity score matching to balance basic demographics. We also compared the presence of scoliosis. Our findings suggest that both lumbar and caudal epidural analgesia can be safely used in OI patients. In cases where lumbar punctures may pose challenges due to potential spine deformities, the caudal route can be an alternative.

Keywords: Lumbar epidural, caudal epidural, osteotomy surgery, osteogenesis imperfecta, cohort analysis, pain management

Introduction

Osteogenesis imperfecta (OI) is a generalized connective tissue disorder resulting in skeletal deformity and susceptibility to fractures in long bones, occurring in 1 in 10,000–20,000 births.¹ Patients with OI may also have comorbidity problems like scoliosis, vertebral compression, impairment of pulmonary function, and cardiac valve abnormalities.² Disease management options, such as physical rehabilitation, pharmacological treatments, and surgical interventions, are chosen depending on the specific type of OI.^{3,4} Patients with OI commonly present with multiple fractures or deformities in their extremities or spines.^{5,6} Orthopedic management often involves realignment through osteotomies in both the upper and lower extremities as well as the spine.^{4,7} These patients may undergo several traumatic surgical procedures, each with a prolonged duration. Furthermore, patients with OI may suffer chronic pain across all types in their lives.⁸ It is crucial to ensure appropriate analgesic measures for these patients, especially in children.⁹ While opioids can be used for pain relief during the perioperative period, their action on various receptors throughout the body can lead to opioid-related adverse effects such as nausea, vomiting, constipation, and respiratory depression¹⁰ and lead to poorly controlled pain.¹¹

Epidural analgesia can provide highly targeted pain relief by delivering analgesic agents directly to the neural structures within the spinal cord. This reduces the need for opioids, thereby mitigating opioid-related nausea, vomiting, respiratory depression, and constipation.^{12,13} Additionally, it helps attenuate the physiological stress response elicited by surgical interventions, resulting in more stable hemodynamics. Furthermore, epidural analgesia has been associated with a decreased incidence of chronic post-surgical pain, decreased the general medication dosage, shorter hospitalization duration, and lower postoperative mortality rates.^{14,15}

Continuous epidural infusion has been empirically validated as an effective and safe technique for providing optimal anesthesia and analgesia in pediatric populations.^{16–18} While lumbar epidural remains a common technique, caudal blocks are also documented as effective strategies for young pediatric patients for interventions below the umbilical region, such as congenital diaphragmatic hernia (CDH).¹⁹ In addition, although single-shot techniques are more common in pediatric cases, catheter placement is easier in this population.^{20,21} Both techniques, when performed in major sub-umbilical surgical procedures, provide continuous analgesia intraoperatively and postoperatively.

Patients with OI may exhibit brittle bones and also spine manifestations, including vertebral rotation and spinal stenosis,²² which can make lumbar puncture procedures more challenging. However, previous studies have shown the successful use of epidural and neuraxial analgesia in such cases.²³ In addition, evidence regarding whether lumbar epidural or caudal epidural provides better pain control for osteotomy surgeries in OI patients with combined spine deformity remains limited.

The University of Hong Kong-Shenzhen Hospital (HKU-SZH) is a major referral center for OI patients in southern China, supported by the Rare Diseases Public Welfare Organization. At our center, both caudal and lumbar epidural analgesia techniques have been administered to pediatric OI patients. This retrospective study aimed to compare the pain score, puncture-related parameters, oral medication regimens, and potential complications between the lumbar and caudal epidural analgesia techniques to determine which one is superior.

Materials and Methods

This retrospective study was conducted at the Hong Kong University- Shenzhen Hospital, a renowned center of surgical care to patients with OI since 2015. This study complies with the Declaration of Helsinki. Ethics approval and institutional review board (IRB hkuszh2023065) were diligently obtained for this study, meanwhile, an exemption for informed consent has been submitted to and approved by the Ethics Committee. The data collection involved electronic medical records and perioperative analgesia paper charts. All data were confirmed by the second reviewer to ensure data accuracy and reliability.

Participant Enrollment

A total of 349 cases less than 1–17 years old with OI who underwent lower extremities osteotomy surgeries with epidural analgesia (lumbar or caudal route) postoperative between 2015–2023 were reviewed. Exclusion criteria included patients with plaster fixation, internal fixation removal, patients who use PCA for analgesia, and cases that lack analgesia records. 274 cases were selected and divided into two groups: Lumbar epidural (Group L) and Caudal (Group C). A 1:1 Propensity score matching (PSM) was used to balance patients' basic characteristics including age, height, and weight. Finally, 90 cases were determined with 45 in each group (Figure 1).

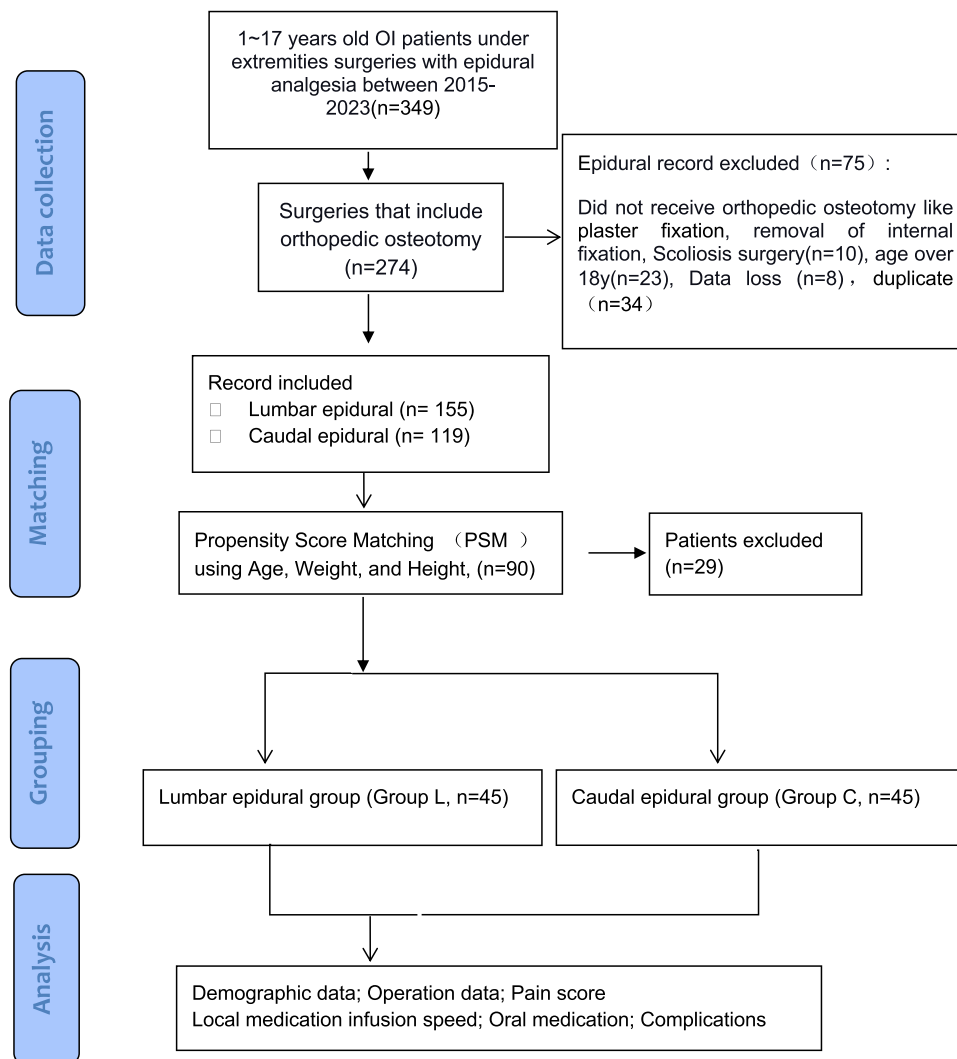


Figure 1 Study flowchart.

Anesthesia and Analgesia

All patients were administered general anesthesia with total intravenous anesthesia (TIVA). Sevoflurane anesthesia was only briefly utilized in pediatric patients who were uncooperative with intravenous cannulation. Propofol, cisatracurium or rocuronium, and fentanyl were used for induction. Following the secure of the airway, maintenance was achieved using propofol and remifentanyl. Patients were gently positioned laterally, with the lumbar or caudal epidural chosen according to the patient's spine scoliosis degree and height. Morphine or sufentanil was supplemented as necessary.

Caudal Epidural Technique

An equilateral triangle was outlined by palpating and aligning the posterior superior iliac spines, with the apex indicating the sacral hiatus position, covered by a sacrococcygeal membrane. An 18 or 16 G IV catheter (Becton, Dickinson Company, BD Angiocath IV catheter, Sandy, Utah, USA) was inserted at a 30–40-degree angle from the midpoint of the line connecting the sacral cornua. Confirmation of membrane penetration was noted as a “loss of resistance”, followed by slight needle flattening and cannula advancement into the caudal space. A 4–5cm epidural catheter was inserted into the caudal space, creating a 3cm tunnel with a 50 mL syringe needle or epidural needle. The catheter was repeatedly aspirated to confirm the absence of blood and cerebrospinal fluid. 3 to 5 mL of 0.5% ropivacaine was administered, followed by a loading dose of 0.5 mL/kg of local anesthetic. Hemodynamics were closely monitored throughout the procedure.

Lumbar Epidural Technique

After the site of L1-L4 interspace was located, either a midline or paramedian epidural insertion procedure was employed with a 17G or 20G epidural needle. Ultrasound guidance was used when facing puncture difficulty or depending on the anesthetists' habits. Approximately 4–5 cm of the catheter was advanced into the epidural space. The catheter was repeatedly aspirated to ensure the absence of blood and cerebrospinal fluid. Subsequently, 3 to 5 mL of 0.5% ropivacaine was administered, followed by a loading dose of 5mL local anesthetic. Hemodynamic parameters were continuously monitored throughout the procedure. The catheter was fixed with a 3M Tegaderm transparent dressing and 3M tape. Both groups of patients were administered with 0.15% to 0.2% ropivacaine infusions during surgery.

Postoperative Analgesia

Postoperative analgesia was maintained through a continuous infusion of 0.1% to 0.125% ropivacaine. The initial infusion rate was set at 4–8 mL/h and adjusted based on the patient's pain intensity. Regular oral medications, such as paracetamol and ibuprofen, were prescribed in most cases after surgery. Low-dose tramadol was administered in instances where inadequate analgesia was observed.

Follow-Up

The acute pain team, consisting of the acute pain service attending and a nurse practitioner, visited the patient on the morning following the surgery. Numeric Rating Scale (NRS) scores or the Face, Legs, Activity, Cry, Consolability (FLACC) scale were employed to assess patient pain based on their age and cognitive capabilities. Oral medication or local infusion rate was adjusted as needed. To mitigate infection risk, epidural catheters are limited to a maximum use of four days. The pain management service concluded the following day after the removal of the epidural catheter. Pain scores and any complications were documented on paper charts, which were later entered into the computer by the pain nurse.

Outcomes

The primary outcome focused on the difference in daily pain severity distribution between the lumbar epidural group and the caudal epidural group after surgery. Secondary outcomes included catheter depth, catheter placement in the epidural space, puncture duration time, and local analgesia infusion rate. Furthermore, we compared oral medication and complications such as back pain, urinary retention, nausea, vomiting, and nerve-related complications between the two groups.

Statistical Analysis

Data were described as mean with standard deviation including 95% confidence intervals (CIs), median (interquartile range), frequency, and percentages when appropriate. Propensity score matching was employed with a 1:1 ratio and a 0.1 tendency score to emulate randomization. Pain scores were categorized as mild, moderate, or severe. Descriptive statistics were used to analyze complications, and an independent sample *t*-test was applied for normally distributed data. The Mann–Whitney *U*-test was selected for non-normally distributed and ordinal data. To compare categorical data, the Chi-Square test or Fisher's exact tests were performed. A *p*-value of less than 0.05 was considered statistically significant. Statistical analysis was conducted using IBM Statistics 26.0 (SPSS, Inc., Chicago, IL, USA).

Results

All 274 cases with 155 lumbar and 119 caudal epidurals were included in this study. Before propensity score matching, the caudal group was younger with lower body weight and height. In addition, the operation time and blood loss were not equal between the two groups before matching. Age, height, and weight were balanced after propensity score matching with 45 cases for each group. The mean age in the lumbar epidural group (Group L) was 7.75 years (range 2–17), while in the caudal epidural group (Group C) was 7.61 years (range 1–15). Additionally, the mean height for the two groups was 117.73 (range 78–167) and 97.51 (range 55–126), respectively. The body weight of the two groups was 26.91 (range 10.5–60) and 17.890 (range 8–42), respectively. Additionally, there were no significant differences in gender, operation time, operation site, and blood loss between the two groups. Furthermore, there were no differences in scoliosis distribution pre-match and after-match. (Table 1).

Primary Outcome - Daily Pain Score

The pain scores were categorized into three grades: 0–3 for mild pain, 4–6 for moderate pain, and 7–10 for severe pain. Notably, the percentage of moderate to severe pain during movement was significantly higher in Group L compared to Group C on Day 2, with rates of 62.5% versus 82.5% for mild pain, 32.5% versus 17.5% for moderate pain, and 5% versus 0% for severe pain during movement ($P=0.039$). However, no statistically significant differences in pain severity distribution were observed between the two groups on Day 1, Day 2, and Day 3 at rest (Table 2).

Secondary Outcomes

The caudal epidural group (Group C) exhibited a greater depth of catheter insertion in the epidural space, with a median 5cm compared to the Lumbar epidural group (Group L) with a median insertion depth of 4cm ($P=0.000$). There were no statistical differences in the depth of catheter insertion from skin to epidural space. The median puncture time in the caudal epidural group was 20(17) minutes, which was longer than the lumbar epidural group, which had a median of 15(9) minutes ($P=0.036$).

There were no statistically significant differences observed between the two groups in terms of postoperative local anesthetic infusion rates. Specifically, the mean infusion rate on day 1 for Group L was 0.321 ± 0.127 mg/kg/h, whereas for Group C, it was 0.316 ± 0.114 mg/kg/h ($P=0.823$). In addition, there was also no significant difference on day 2, ($P=0.214$) and day 3 ($P=0.849$) between the two groups. In the days of catheter use distribution, there was also no significant difference between the two groups, ($P=0.360$) (Table 3). No infection was observed in both groups.

Both groups were prescribed oral medications after the operation. There was no statistical difference in oral medication taken between the two groups on Day 1. On the first day, 73.7% of patients in the lumbar epidural group used paracetamol, while 68.9% in the caudal epidural ($P=0.642$). NSAIDs were used by 68.9% of patients in the lumbar epidural group and 84.4% in the caudal epidural group ($P=0.081$) respectively. On the other hand, tramadol was used by 26.7% of patients in the lumbar epidural group and 15.6% in the caudal epidural group ($P=0.197$). In addition, there were no significant differences in the oral medications taken between the two groups on Day 2 and Day 3 (Table 4).

There was no significant difference in postoperative complications between the two groups. In the postoperative period, one case (2.22%) of back pain was found in each group. One case (2.22%) of urinary retention was observed in the caudal group. In the lumbar epidural group, two cases (4.44%) exhibited symptoms of nausea and vomiting while three cases (6.67%) in the caudal group. No nerve injury or numbness was observed in both groups (Table 5).

Table 1 Baseline Characteristics

Baseline Characteristics		Before PSM			After PSM		
		Lumbar (n=155)	Caudal (n=119)	P	Lumbar (n=45)	Caudal (n=45)	P
Mean Age (SD), y		10.29±3.3, (n=151)	5.56 ±3.43, (n=118)	0.000*	7.75±2.75, (n=45)	7.61±2.95, (n=45)	0.747
Mean Height (SD), cm		117.73±19, (n=151)	97.51±16.34, (n=118)	0.000*	106.31±14.789, (n=45)	104.47±13.10, (n=45)	0.833
Mean Weight (SD)kg		26.91±9.57, (n=151)	14.59±5.41, (n=118)	0.000*	19.18±5.79, (n=45)	18.14±6.08, (n=45)	0.220
Gender, n(%)							
	Male	98/155, (63.2%)	69/119, (58%)	0.378	27/45, (60%)	25/45, (53.3%)	0.523
	Female	57/155, (63.2%)	50/119, (42%)		18/45, (40%)	21/45, (46.7%)	
Operation Time (Median (IQR))		319(226), (n=149)	257.5(168), (n=102)	0.038*	301(192), (n=45)	260(170), (n=45)	0.787
Operation Site				0.802	34(50%)	35(50%)	1.000
	Upper the Knee	104(72.2%)	81(73.6%)				
	Below the Knee	40(27.8%)	29(26.4%)				
Blood Loss (Median (IQR)), mL		200(250), (n=149)	100(150), (n=102)	0.000*	150(175), (n=45)	100(190), (n=45)	0.283
Scoliosis yes		61(42.67%)	33(29.46%)	0.079	33(80.5%)	31(79.6%)	0.566
	No	82	79		8	8	
	No Record	12	7		4	6	

Note: *p<0.05 is considered statistically significant.

Abbreviation: PSM, propensity score matching.

Table 2 Comparison of Daily Pain Scores Between the Lumbar and Caudal Groups

Pain scores	Grade	Day 1						Day 2						Day 3					
		Rest			Movement			Rest			Movement			Rest			Movement		
		L(n=45)	C(n=45)	P	L(n=40)	C(n=40)	P	L(n=40)	C(n=40)	P	L(n=40)	C(n=40)	P	L(n=27)	C(n=31)	P	L(n=27)	C(n=31)	P
0~3	1	37	42	0.101	19	27	0.051	39	40	0.317	25	33	0.039*	27	31	1.000	23	37	0.875
		82.22%	93.33%		42.20%	60.00%		97.50%	100%		62.50%	82.50%		100%	100%		85.19%	87.10%	
4~6	2	6	3		19	16		1	0		13	7					3	3	
		3.33%	6.67%		42.22%	35.56%		2.50%	0		32.50%	17.50%					11.11%	9.68%	
7~10	3	2	0		7	2					2	0					1	1	
		4.44%	0		15.56%	4.44%					5.00%	0					3.70%	3.23%	

Note: *p<0.05 is considered statistically significant.

Abbreviations: L, Lumbar; C, Caudal.

Table 3 Puncture Parameters and Infusion Speed After Operation (Mean ± SD)

Second outcomes	Group L (N=45)	Group C (N=45)	P-value
	Mean ±SD (n)	Mean± SD (n)	
Catheter in Epidural, cm	4(1.8),(n=45)	5(3),(n=41)	0.000*
Needle Depth, cm	2.5(1),(n=45)	3(1),(n=39)	0.700
Puncture Duration, min	15(9),(n=45)	20(17),(n=45)	0.036*
Infusion Speed, mg/kg/h			
Day 1	0.321±0.127(n=44)	0.316±0.114(n=44)	0.823
Day 2	0.298±0.119(n=40)	0.3209±0.104(n=41)	0.214
Day 3	0.303± 0.104(n=27)	0.308±0.098(n=31)	0.849
Patient Count by Catheter Use Days			
1 Day	4	3	0.360
2 Day	13	11	
3 Day	12	20	
4 Day	16	11	

Note: *p<0.05 is considered statistically significant.

Abbreviations: L, Lumbar; C, Caudal.

Table 4 Number of Oral Medications Post-Operatively Taken Post-Operatively (Lumbar Epidural, L, n=45; Caudal Epidural, C, n=45)

	Day 1		p-value	Day 2		p-value	Day 3		p-value
	Group L	Group C		Group L	Group C		Group L	Group C	
Paracetamol	33(73.3%)	31(68.9%)	0.642	25(55.6%)	30(66.7%)	0.280	15(33.3%)	21(46.7%)	0.197
NSAID	31(68.9%)	38(84.4%)	0.081	27(60%)	32(71.1%)	0.267	16(35.6%)	25(55.6%)	0.057
Tramadol	12(26.7%)	7(15.6%)	0.197	8(17.8%)	6(13.3%)	0.561	8(17.8%)	5(11.1%)	0.368

Note: p<0.05 is considered statistically significant.

Abbreviations: L, Lumbar; C, Caudal.

Table 5 Postoperative Adverse Effects and Complications

Catheter and complications	Group L	Group C	P
	n=45	n=45	
Back Pain	1(2.22%)	1(2.22%)	0.753
Urinary Retention	0	1(2.22%)	0.500
Nausea	2(4.44%)	3(6.67%)	0.500
Vomiting	2(4.44%)	3(6.67%)	0.500
Nerve Injury-Related Problems	0	0	

Note: p<0.05 is considered statistically significant.

Abbreviations: L, Lumbar; C, Caudal.

Discussion

OI is an uncommon, progressive skeletal disorder that leads to severe skeletal deformities, making individuals susceptible to fractures.⁵ This condition often requires multiple surgical interventions, especially during the critical growth stages of their skeletal development. Managing pain during the perioperative period of pediatric orthopedic surgery, especially in patients undergoing multiple osteotomies, can be a challenging task. The multimodal analgesia approach has benefits in reducing opioid dosages and opioid-related side effects like respiratory depression, nausea, vomiting, and hyperalgesia during the perioperative period.¹³ Regional analgesia combined with other mechanism medications should be considered for long-duration and complex surgeries, such as those studied here, where the median operation time is four to five hours. Epidural anesthesia not only provides better analgesia in and after the operation but also decreases opioid exposure perioperatively.¹⁵ While epidural analgesia was proven to be used in pediatric surgeries, caudal epidural can also be used in lower extremity surgeries.¹⁹

For OI patients, factors such as bone fragility, deformity, and sometimes the requirement for plaster immobilization make the process of puncture particularly challenging. Previous research shows neuraxial analgesia is successfully used in OI patients.²³ While in our hospital, a local OI center, the epidural was usefully used for seven years without any nerve-related complications. In our center, the epidural was typically administered after general anesthesia in most children. Lumbar epidural or Caudal epidural was used in OI patients depending on the patient's height and spine deformities, as well as the curvature of the spine and the spinous process. To compare the two methods with more precision, we employed propensity score match (PSM) to balance the basic characteristics such as age, height, and weight. After applying PSM, no differences were observed in terms of operation time, blood loss, daily ropivacaine infusion speed, and oral medication between the two groups.

The reason why we divided pain scores into three degrees (mild, 0–3; moderate, 4–6; severe, 7–10) was that we found both groups had pain scores of ten and it is hard to decide whether the catheter location was good as it is a retrospective study. This extreme value may influence the median pain score value if we direct inclusion in statistics and, it is not statistical to exclude the extremes. After analyzing pain scores according to the pain degree, the caudal epidural showed a lower pain score distribution during movement on the second day. The reason why there was no difference on the first day may be attributed to that most surgeries finished late in the day, and there still was some systemic analgesia effect and a high concentration of local analgesia in the epidural on the subsequent day. Additionally, adjustments to oral medication and catheter placement were made in response to pain scores and sensory levels, together with a potential decrease in pain stress by the third day, which could explain our inability to detect a distinction on the third day.

Furthermore, our findings suggest that a caudal epidural can be an effective analgesia option even during surgeries at higher anatomical levels in pediatric patients, 75.56% of cases (34/45) were performed with the operation above the knee. On the other hand, scoliosis is a prevalent comorbid disease in individuals with OI patients, contributing to spine deterioration and exhibiting associations with various genetic factors, age, bone mineral density. In a study conducted by Pei Kai Chen, the research indicated that with each advancing year, there is an incremental odds ratio of 1.13 (95% CI, 1.07–1.2) for progression into advanced stages of scoliosis, as determined through multivariate logistic regression analysis.²⁴ That may remind us of the difficulty of puncture with the age growing. According to Amy R. Beethe's review study, 161/117(72.67%) of patients had used lumbar epidural analgesia in this rare disease, but only 7/161(5.9%) of those patients reported using caudal analgesia.²³ Our results show lumbar epidural were used in 61/143(80.5%) OI patients combined with scoliosis while caudal epidural was used in 31/39(79.6%) with scoliosis. Though the lumbar epidural route remains a conventional approach for pediatric postoperative pain control management, the caudal epidural is also a viable alternative for pediatric OI patients,¹⁹ particularly those with OI disease combined with scoliosis, lumbar route failed. Our research shows caudal route requires longer catheter insertion, (5(3) cm in the caudal group versus 4 (1.8) cm in the lumbar group), despite this, the relatively stable anatomy of the caudal region may provide an advantage in maintaining the catheter in central position of the spinal canal in OI patients with vertebral contortion. On the other hand, earlier research has indicated that severe forms of OI can result in spinal deformities, which in turn may lead to a decrease in trunk height,²⁵ which may explain the efficacy of caudal epidural at higher anatomical operation sites. Ultrasound can be employed to locate the catheter tip for further study.^{26,27}

In our study, as part of a multimodal approach for this traumatic surgery, paracetamol and ibuprofen^{28,29} were prescribed to most of the patients, if pain control was still not enough or breakthrough pain still existed, tramadol orally

or intramuscular was used. There is no difference between the two groups for the medications taken, and no difference between tramadol uses in subgroup analysis. It is better for moderate to severe pain, especially for surgeries that impact peripheral nerves. Though tramadol was warned against use under 12 years by FDA black boxes, it still provides good analgesia for neuropathic pain. In our research, 18/19(94.7%) tramadol was used over six years old under a lower dosage without respiratory depression and the epidural analgesia also had an opioid-sparing effect which decreased the respiratory depression incidence after the operation. We also have a close monitoring protocol that includes monitoring consciousness and respiratory rate, as well as a rescue regimen in the ward.

Epidural analgesia effectiveness can be influenced by numerous factors, such as the catheter type, duration of the procedure, surgical site, and oral medications, among others. To obtain an acceptable sample size, our statistical analysis adjusted three basic characteristics such as age, height, and weight. Fortunately, after PSM, other factors like operation time, operation site, and oral medication were comparable.

Limitation

This study is retrospective in nature, and treatment selection was not randomized. Although we employed propensity score matching to minimize bias, it resulted in the loss of many cases. As with other studies on rare diseases, the sample size in this research may be insufficient. In our clinical practice, we conduct ward rounds once daily and record morning pain scores. Future studies should document pain score distributions more comprehensively throughout the day. Severe pain incidents were managed by the on-call doctor at night through adjustments to catheter depth or infusion rate, but this information was not adequately recorded in our documentation.

Furthermore, genotyping, the degree of scoliosis, and the level of puncture difficulty were not thoroughly recorded and compared. Ultrasound and X-rays should be used in the future to investigate the catheter tip position. Future studies should also investigate long-term analgesia, the use of opioids in epidural procedures, the best drugs to employ and their dosage, the criteria to choose one of the two approaches, sleep quality, and psychological factors. Data entry in our current database is manual, and we are working to establish an electronic record system for more efficient and accurate data management. A randomized controlled trial with a well-defined design and more comprehensive data collection will be necessary in future research.

Conclusion

Both lumbar and caudal epidural techniques can effectively alleviate pain after lower extremity osteotomy surgery in children with osteogenesis imperfecta. When there is significant spinal deformity and rotation, ultrasound should be available to help the anesthesia provider to locate the targeted intervertebral level in a difficult lumbar epidural puncture. If it is challenging, a caudal epidural may be an alternative option.

Data Sharing Statement

Due to privacy concerns, data from this paper will not be made publicly available. However, anonymized data may be shared upon reasonable request to the corresponding authors.

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Disclosure

The authors have no conflicts of interest to declare in this work.

References

1. Deguchi M, Tsuji S, Katsura D, Kasahara K, Kimura F, Murakami T. Current overview of osteogenesis imperfecta. *Medicina*. 2021;57(5):464. doi:10.3390/medicina57050464
2. Marom R, Rabenhorst BM, Morello R. Osteogenesis imperfecta: an update on clinical features and therapies. *Eur J Endocrinol*. 2020;183(4):R95–R106. doi:10.1530/EJE-20-0299
3. Palomo T, Vilaça T, Lazaretti-Castro M. Osteogenesis imperfecta: diagnosis and treatment. *Curr Opin Endocrinol Diabetes Obes*. 2017;24(6):381–388. doi:10.1097/MED.0000000000000367
4. Perea SH, Green DW. Osteogenesis imperfecta: treatment and surgical management. *Curr Opin Pediatr*. 2021;33(1):74–78. doi:10.1097/MOP.0000000000000968
5. Forlino A, Marini JC. Osteogenesis imperfecta. *Lancet*. 2016;387(10028):1657–1671. doi:10.1016/S0140-6736(15)00728-X
6. Jovanovic M, Guterman-Ram G, Marini JC. Osteogenesis imperfecta: mechanisms and signaling pathways connecting classical and rare OI types. *Endocr Rev*. 2022;43(1):61–90. doi:10.1210/endrev/bnab017
7. Marini JC, Forlino A, Bächinger HP, et al. Osteogenesis imperfecta. *Nature Reviews Disease Primers*. 2017;3(1):17052. doi:10.1038/nrdp.2017.52
8. Rodriguez Celin M, Kruger KM, Caudill A, et al. A multicenter study to evaluate pain characteristics in osteogenesis imperfecta. *Am J Med Genet A*. 2023;191(1):160–172. doi:10.1002/ajmg.a.63009
9. Celin MR, Kruger KM, Caudill A, et al. A multicenter study to evaluate pain characteristics in osteogenesis imperfecta. *Am J Med Genet A*. 2023;191(1):160–172. doi:10.1002/ajmg.a.63009
10. Frauenknecht J, Kirkham KR, Jacot-Guillarmod A, Albrecht E. Analgesic impact of intra-operative opioids vs. opioid-free anaesthesia: a systematic review and meta-analysis. *Anaesthesia*. 2019;74(5):651–662. doi:10.1111/anae.14582
11. Gan TJ. Poorly controlled postoperative pain: prevalence, consequences, and prevention. *J Pain Res*. 2017;10:2287–2298. doi:10.2147/JPR.S144066
12. Heo MH, Kim JY, Kim JH, et al. Epidural analgesia versus intravenous analgesia after minimally invasive repair of pectus excavatum in pediatric patients: a systematic review and meta-analysis. *Korean J Anesthesiol*. 2021;74(5):449–458. doi:10.4097/kja.21133
13. Colvin LA, Bull F, Hales TG. Perioperative opioid analgesia—when is enough too much? A review of opioid-induced tolerance and hyperalgesia. *Lancet*. 2019;393(10180):1558–1568. doi:10.1016/S0140-6736(19)30430-1
14. Pöpping DM, Elia N, Van Aken HK, et al. Impact of epidural analgesia on mortality and morbidity after surgery: systematic review and meta-analysis of randomized controlled trials. *Ann Surg*. 2014;259(6):1056–1067. doi:10.1097/SLA.0000000000000237
15. Heydinger G, Tobias J, Veneziano G. Fundamentals and innovations in regional anaesthesia for infants and children. *Anaesthesia*. 2021;76(Suppl 1):74–88. doi:10.1111/anae.15283
16. Gupta A, Jay MA, Williams G. Evolving pediatric epidural practice: an institution’s clinical experience over 20 years—A retrospective observational cohort study. *Paediatr Anaesth*. 2020;30(1):25–33. doi:10.1111/pan.13767
17. Bravenboer-Monster K, Keyzer-Dekker C, van Dijk M, Staals L, Leeuw T, Wijnen R. Efficacy of epidural analgesia after laparotomy in children. *Eur J Pediatr Surg*. 2019;29(2):209–214. doi:10.1055/s-0038-1632372
18. Warmann SW, Lang S, Fidele F, et al. Perioperative epidural analgesia in children undergoing major abdominal tumor surgery—A single center experience. *J Pediatr Surg*. 2014;49(4):551–555. doi:10.1016/j.jpedsurg.2013.10.025
19. Wiegele M, Marhofer P, Lönnqvist P-A. Caudal epidural blocks in paediatric patients: a review and practical considerations. *Br J Anaesth*. 2019;122(4):509–517. doi:10.1016/j.bja.2018.11.030
20. Sanghvi C, Dua A. *Caudal Anesthesia*. In: *StatPearls*. Treasure Island (FL): StatPearls Publishing; 2023.
21. Soaida SM, ElSheemy MS, Shouman AM, et al. Caudal extradural catheterization in pediatric renal transplant and its effect on perioperative hemodynamics and pain scoring: a prospective randomized study. *J Anesth*. 2016;30(1):47–54. doi:10.1007/s00540-015-2087-8
22. Wallace MJ, Kruse RW, Shah SA. The spine in patients with osteogenesis imperfecta. *J Am Acad Orthop Surg*. 2017;25(2):100–109. doi:10.5435/JAAOS-D-15-00169
23. Beethe AR, Bohannon NA, Ogun OA, et al. Neuraxial and regional anesthesia in surgical patients with osteogenesis imperfecta: a narrative review of literature. *Reg Anesth Pain Med*. 2020;45(12):993–999. doi:10.1136/rapm-2020-101576
24. Chen P, Zhou Y, Tan Z, et al. Scoliosis in osteogenesis imperfecta: identifying the genetic and non-genetic factors affecting severity and progression from longitudinal data of 290 patients. *Orphanet J Rare Dis*. 2023;18(1):295. doi:10.1186/s13023-023-02906-z
25. Jain M, Tam A, Shapiro JR, et al. Growth characteristics in individuals with osteogenesis imperfecta in North America: results from a multicenter study. *Genet Med*. 2019;21(2):275–283. doi:10.1038/s41436-018-0045-1
26. Ahiskalioglu A, Yayik AM, Ahiskalioglu EO, et al. Ultrasound-guided versus conventional injection for caudal block in children: a prospective randomized clinical study. *J Clin Anesth*. 2018;44:91–96. doi:10.1016/j.jclinane.2017.11.011
27. Kil HK. Caudal and epidural blocks in infants and small children: historical perspective and ultrasound-guided approaches. *Korean J Anesthesiol*. 2018;71(6):430–439. doi:10.4097/kja.d.18.00109
28. Krauss BS, Calligaris L, Green SM, Barbi E. Current concepts in management of pain in children in the emergency department. *Lancet*. 2016;387(10013):83–92. doi:10.1016/S0140-6736(14)61686-X
29. O’Neill A, Lirk P. Multimodal Analgesia. *Anesthesiol Clin*. 2022;40(3):455–468. doi:10.1016/j.anclin.2022.04.002

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