

Opioid-Sparing Thoracotomy Analgesia Using an Erector Spinae Plane Block in an Infant with Autosomal Recessive Congenital Ichthyosis: A Case Report

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ABSTRACT

Background: The erector spinae plane (ESP) block is increasingly used for pediatric thoracic analgesia, yet evidence in infants under one year remains limited. Thoracotomy is associated with severe postoperative pain and high opioid requirements. Autosomal recessive congenital ichthyosis (ARCI), characterized by impaired epidermal barrier function, raises concerns regarding the safety of regional anesthesia. No previous report has described the use of regional anesthesia in patients with ARCI.

Case: A 10-month-old, 9.2 kg male infant with genetically confirmed ARCI (PNPLA1 mutation) underwent left posterolateral thoracotomy and decortication for loculated empyema. A pre-incision ultrasound-guided ESP block was performed at T4 using 0.25% bupivacaine (4.5 mL; 1.22 mg/kg). Intraoperative opioid use was limited to fentanyl 25 mcg for intubation, with no additional analgesic requirement or hemodynamic response to surgical incision. Postoperative pain scores assessed using the face, legs, activity, cry, consolability (FLACC) scale remained 1/10 during the first 24 hours, increased to 6/10 on post-operative day (POD)-1, and subsequently decreased to 4/10 on POD-2 and 3/10 on POD-3 under multimodal non-opioid analgesia. No rescue opioids or opioid-related adverse effects occurred. The patient was extubated in the operating room and discharged from the pediatric intensive care unit (PICU) after 58 hours. No block-related complications were observed, and puncture-site healing was uneventful.

Discussion: This case demonstrates that the ESP block provided effective opioid-sparing analgesia in an infant and suggests that ARCI-related epidermal barrier dysfunction did not appear to adversely affect the safety or technical performance of an interfascial plane block in this case.

Conclusion: ESP block appeared to be a feasible, effective, and safe analgesic option for thoracotomy in this infant with ARCI and may serve as an opioid-sparing strategy in high-risk pediatric patients.

Keywords: autosomal recessive congenital ichthyosis; erector spinae plane block; infant; opioid-sparing analgesia; regional anesthesia; thoracotomy

INTRODUCTION

The erector spinae plane (ESP) block has gained increasing acceptance as an ultrasound-guided interfascial plane block for thoracic and upper abdominal analgesia since its first description by Forero et al. in 2016.¹ The technique involves injection of local anesthetic deep to the erector spinae muscle (ESM), allowing cranio-caudal spread with potential involvement of the dorsal and ventral rami and variable paravertebral extension.^{2,3} Its relative technical simplicity and favorable safety profile, given its distance from the pleura and neuraxis, make it an attractive option for thoracic procedures. Thoracotomy is associated with severe postoperative pain and substantial opioid requirements, underscoring the need for effective analgesic strategies, particularly in pediatric patients.⁴

Pediatric applications of the ESP block have expanded in recent years, including use in thoracic, cardiac, and upper abdominal surgery.^{5,6,7} However, evidence in infants, particularly those under one year of age, remains limited. This population presents unique challenges, including narrow safety margins for local anesthetic dosing and increased susceptibility to opioid-related respiratory adverse effects, highlighting the importance of effective opioid-sparing analgesic strategies.⁸

Autosomal recessive congenital ichthyosis (ARCI) is a rare genodermatosis characterized by impaired epidermal differentiation and barrier dysfunction, resulting in generalized scaling and xerosis.⁹ Mutations in genes involved in epidermal lipid metabolism, including PNPLA1¹⁰ and TGM1¹¹, contribute to abnormal skin structure and function. From an anesthetic perspective, ARCI

raises theoretical concerns about the safety of regional anesthesia, including an increased risk of puncture-site infection, altered tissue architecture, and the potential unpredictability of local anesthetic spread.

To date, no previous report has described the use of regional anesthesia in patients with ARCI. This case highlights the feasibility and safety of the ESP block in an infant with ARCI undergoing thoracotomy. It demonstrates its potential as an effective opioid-sparing analgesic strategy in high-risk pediatric patients.

CASE

A 10-month-old, 9.2 kg male infant (weight-for-age within normal range, length 69 cm) with genetically confirmed ARCI due to a PNPLA1 mutation presented with a 2-day history of high-grade fever (peak 39.8°C), cough, rhinorrhea, and post-tussive vomiting. Clinical examination revealed generalized xerosis and diffuse scaling consistent with ARCI.

The patient was born at 36 weeks' gestation via cesarean section for anhydramnios with meconium-stained amniotic fluid and required short-term respiratory support after birth. Developmental milestones were appropriate for age, with mild delay in gross motor function.

Initial management for a suspected upper respiratory tract infection was conservative. Despite treatment, clinical deterioration occurred on hospital day 3, characterized by tachypnea, oxygen desaturation (SpO₂ 89% on room air), and increased work of breathing with intercostal retractions. Chest radiography demonstrated left lower lobe consolidation with pleural effusion.

Laboratory findings showed leukocytosis (11,750/ μ L) and elevated C-reactive protein (53.6 mg/L), and antibiotic therapy was escalated to intravenous ceftazidime.

Thoracic ultrasound confirmed a complex left pleural effusion with septations, consistent with evolving empyema. Blood cultures yielded methicillin-sensitive *Staphylococcus aureus*. Ultrasound-guided thoracostomy was performed, but drainage remained minimal. Despite seven days of targeted antibiotic therapy, the patient exhibited persistent fever and worsening inflammatory markers, with C-reactive protein rising to 124 mg/L.

Contrast-enhanced computed tomography revealed a loculated left-sided empyema with mediastinal extension and compressive atelectasis. Given the failure of medical management and pleural drainage, definitive surgical intervention was indicated.

The patient was scheduled for left posterolateral thoracotomy with empyema evacuation and pleural decortication. Preoperative evaluation showed stable hemodynamics with oxygen supplementation (2 L/min nasal cannula) and a face, legs, activity, cry, consolability (FLACC) score of 1/10. Considering the anticipated severity of post-thoracotomy pain and the need to minimize systemic opioid exposure in an infant, an ultrasound-guided regional anesthesia strategy was planned as part of perioperative analgesic management.

After obtaining informed parental consent, the patient was transferred to the operating room. Standard monitoring according to the American Society of Anesthesiologists (ASA) recommendations was applied, with particular attention to skin

preparation due to underlying ARCI. A central venous catheter (CVC 4.0 Fr, double lumen) had been previously placed in the left femoral vein in the pediatric intensive care unit (PICU) and was used for intraoperative access.

General anesthesia was induced with sevoflurane in oxygen. Fentanyl 25 mcg (2.7 mcg/kg) was administered during induction to facilitate airway manipulation. Adequate mask ventilation was confirmed before airway instrumentation. Neuromuscular blockade was achieved with atracurium 4 mg (0.43 mg/kg).

Following induction, direct laryngoscopy was performed, and a Fogarty catheter was introduced into the airway as a bronchial blocker. A 4.5 mm cuffed endotracheal tube was then inserted and secured at 13 cm. The bronchial blocker was subsequently repositioned and its final placement confirmed under fiberoptic guidance to selectively occlude the left main bronchus, achieving one-lung ventilation of the right lung.

After lung isolation was secured and before surgical incision, the patient was positioned in the right lateral decubitus position for regional anesthesia. An ultrasound-guided ESP block was then performed at the T4 level using a high-frequency linear transducer (5–12 MHz) in a longitudinal parasagittal orientation.

The transverse process and overlying ESM were identified. A 22-gauge, 50 mm echogenic needle (Pajunk) was advanced in-plane in a cranio-caudal direction until bony contact with the transverse process was achieved. The needle was then withdrawn slightly to position the tip within the interfascial plane deep to the ESM.

After repeated negative aspiration, 0.5–1 mL of normal saline was injected to confirm correct plane placement by observing linear separation of the ESM from the transverse process (hydrodissection). Subsequently, 4.5 mL of 0.25% bupivacaine (1.22 mg/kg) was administered incrementally under real-time ultrasound visualization, demonstrating longitudinal craniocaudal spread. (Figure 1)

A left posterolateral thoracotomy was performed through the fifth intercostal space. Intraoperative findings included loculated purulent collections, thickened visceral pleura, and an abscess cavity yielding purulent material. Empyema evacuation and pleural decortication were completed, and a chest tube was placed at the end of surgery. Estimated blood loss was approximately 80 mL. Intraoperative fluid management included 250 mL of balanced crystalloid (Ringer's acetate) and 90 mL of packed red blood cells. The total anesthetic time was 240 minutes. After reversal of neuromuscular blockade and restoration of adequate spontaneous ventilation, the patient was extubated in the operating room and transferred to the PICU. No hemodynamic response to surgical incision was observed following block placement. Additional intraoperative analgesia was not required. The procedure was completed without hemodynamic instability or anesthetic complications. No signs of local anesthetic systemic toxicity (LAST) were observed intraoperatively.

Postoperative pain was assessed using the FLACC scale. Pain scores remained low during the first 24 hours following surgery, with a FLACC score of 1/10 on post-operative day (POD)-0. A peak score of 6/10 was observed on POD-1, corresponding to the expected waning of the single-shot ESP block effect. Pain

subsequently decreased to 4/10 on POD-2 and 3/10 on POD-3 under multimodal non-opioid analgesia. Scheduled paracetamol was administered as baseline analgesia, with ketorolac added on POD-1 according to institutional protocol. No rescue opioid analgesia was required at any point during the perioperative or postoperative period. Total opioid exposure was limited to a single intraoperative fentanyl dose of 25 mcg (2.7 mcg/kg). No opioid-related adverse effects, including respiratory depression, postoperative nausea and vomiting, or pruritus, were observed. This pattern suggests an effective analgesic duration of approximately 24 hours, followed by adequate pain control with non-opioid multimodal therapy.

In the PICU, the patient remained hemodynamically stable without vasopressor support. Oxygen supplementation was gradually weaned from the nasal cannula (2 L/min) to room air. Total PICU length of stay was approximately 58 hours. Postoperative laboratory evaluation showed persistent leukocytosis consistent with an inflammatory response, without clinical evidence of sepsis or organ dysfunction. Antibiotic therapy was de-escalated according to clinical and microbiological findings.

Microbiological and pathological evaluations supported the diagnosis of chronic empyema. Pleural fluid, urine, and pleural tissue cultures remained sterile, while the initial blood culture had yielded methicillin-sensitive *Staphylococcus aureus*, as described above. Histopathological examination confirmed chronic empyema with fibrosis. A transient postoperative febrile episode (peak 38.2°C) occurred on POD-1 and resolved without escalation of antimicrobial therapy.

The subsequent clinical recovery was uneventful. Respiratory status improved progressively, allowing full oxygen independence by POD-2. Chest tube output decreased steadily, and the drain was removed on POD-6. The patient was discharged on POD-10 in good clinical condition.

Given the underlying ARCI and the theoretical concern regarding skin barrier dysfunction, the ESP block puncture site was inspected daily. No signs of erythema, discharge, hematoma, or local infection were observed. No clinical features suggestive of LAST occurred. Puncture-site healing was uneventful despite underlying ARCI.

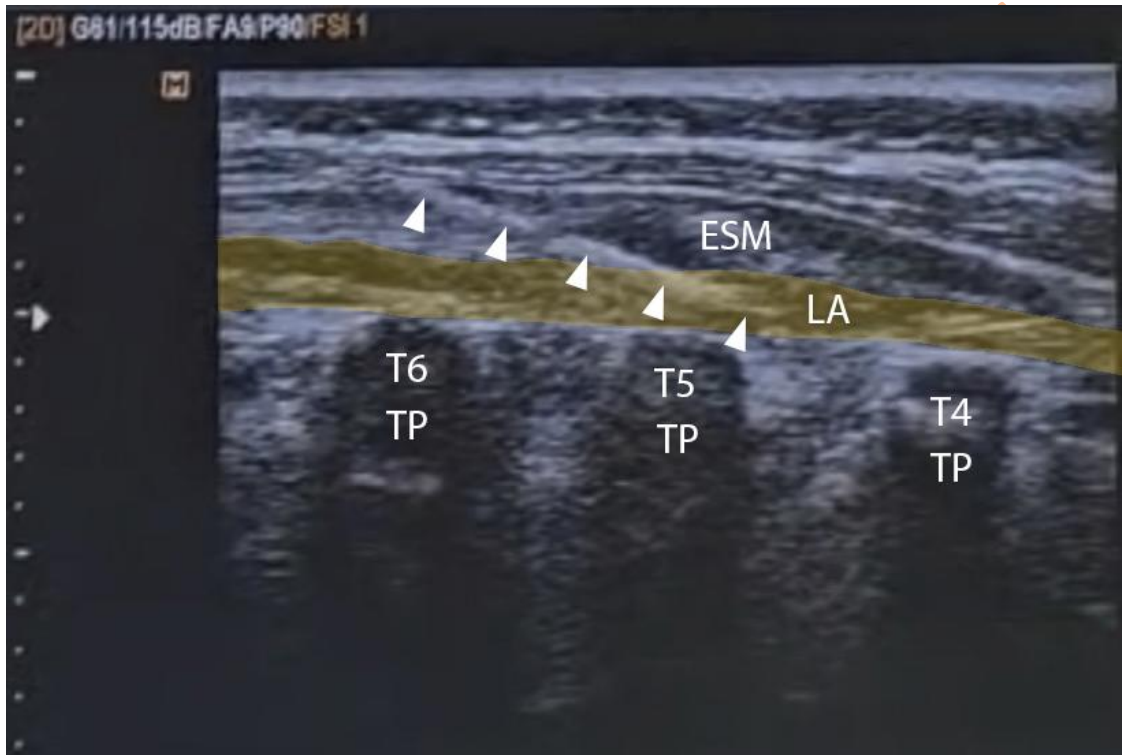


Figure 1. Ultrasound image demonstrating the erector spinae plane (ESP) block at the thoracic level. The transverse processes of T4–T6 (TP) are identified with acoustic shadowing, and the erector spinae muscle (ESM) is visualized superficial to the transverse processes. The needle trajectory is indicated by arrowheads, while local anesthetic spread is observed within the ESP as a hypoechoic layer lifting the muscle from the transverse processes.

DISCUSSION

This report describes a previously unreported use of an ultrasound-guided ESP block for thoracotomy in an infant with ARCI. The novelty of this report lies in the combined context of infant thoracic surgery and a rare disorder of epidermal barrier dysfunction, both of which raise concerns regarding the safety and feasibility of regional anesthesia.

The use of the ESP block in infants remains relatively limited in the literature. Although several studies have demonstrated its efficacy in pediatric thoracic and abdominal surgery, data in patients under one year of age are scarce.^{12,13,14} In the present case, a single-shot ESP block using bupivacaine 0.25% at 1.22 mg/kg provided effective analgesia for approximately 24 hours, as

reflected by low FLACC scores during the first POD. The subsequent increase in pain scores on POD-1, followed by improvement with non-opioid multimodal analgesia, is consistent with the expected pharmacological duration of single-shot local anesthetics in pediatric patients.¹⁵ The effectiveness of the ESP block in this case may be explained by its ability to facilitate cranio-caudal spread of local anesthetic with potential extension into the paravertebral space, providing both somatic and visceral analgesia.¹

The opioid-sparing effect observed in this case is clinically significant. Despite undergoing thoracotomy, which is typically associated with severe postoperative pain and high opioid requirements,¹⁶ the patient required no postoperative opioids. Minimizing opioid exposure in infants is particularly important given their increased susceptibility to respiratory depression and other adverse effects.¹⁷ The absence of intraoperative hemodynamic response to incision further supports the effectiveness of ESP block as a primary regional analgesic technique within a multimodal strategy.

The presence of ARCI introduces additional considerations. Impaired epidermal barrier function raises theoretical concerns regarding puncture-site infection and altered superficial tissue integrity.¹⁸ However, no technical difficulty was encountered during block performance, and ultrasound visualization of the interfascial plane was preserved. Furthermore, no local or systemic complications were observed, and puncture-site healing was uneventful. These findings suggest that ARCI-related epidermal barrier dysfunction did not adversely affect ESP block performance or puncture-site

healing in this case. This observation challenges the traditional concern that impaired skin barrier function may increase procedural risk during regional anesthesia.

When compared with alternative regional techniques for pediatric thoracotomy, the ESP block offers several practical advantages. Thoracic epidural analgesia provides effective pain control but carries risks of neuraxial complications and may be technically challenging in infants.¹⁹ Paravertebral block, while effective, is associated with a higher risk of pleural puncture and may require multiple injections.²⁰ Intercostal nerve blocks have a limited duration and require repeated administration.²¹ In contrast, the ESP block allows multilevel analgesia with a single injection, with a favorable safety profile due to its distance from critical structures.²²

Several limitations should be acknowledged. This report represents a single case, limiting generalizability. Sensory block distribution could not be assessed due to general anesthesia. Pain assessment using the FLACC scale may be subject to observer variability. Additionally, the use of multimodal analgesia may confound the isolated effect of the ESP block, although the absence of opioid requirement supports its significant contribution.

Overall, this case supports the feasibility, safety, and analgesic effectiveness of ESP block in infants, including those with rare dermatologic conditions such as ARCI. The findings expand the potential applicability of interfascial plane blocks and reinforce their role as opioid-sparing strategies in high-risk pediatric populations. Taken together, this case suggests that carefully performed ultrasound-guided

interfascial plane blocks may remain a viable option even in patients with dermatologic conditions traditionally considered at higher risk.

CONCLUSION

Ultrasound-guided ESP block is a feasible, effective, and safe analgesic technique for thoracotomy in infants with ARCI. Epidermal barrier dysfunction did not appear to adversely affect block performance or safety in this case. ESP block may be considered an opioid-sparing component of multimodal analgesia in high-risk pediatric patients.

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