

Anterior rectus sheath blocks in children with abdominal wall pain due to anterior cutaneous nerve entrapment syndrome: a prospective case series of 85 children

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ABSTRACT

Background: Chronic abdominal pain in children may be caused by the anterior cutaneous nerve entrapment syndrome. Local nerve blocks are recommended as an initial treatment in adults. Evidence on effectiveness and safety of such a treatment in children is lacking.

Aim: Our aim was to study outcome and adverse events of anterior rectus sheath blocks in childhood anterior cutaneous nerve entrapment syndrome. **Methods:** Patients <18 years of age receiving anterior rectus sheath blocks were prospectively followed. Injections were administered using a free-hand technique in the outpatient department.

Results: A total of 85 children were included (median age 15 years, range 8–17, 76% female). Eighty-three children reported immediate pain relief following a single lidocaine block and 13 achieved long-term success. Another 19 children was successfully treated with additional blocks combined with steroids. A total 38% success ratio was attained after a median 17-month follow-up (range, 4–39). Pain intensity and diagnostic delay were not associated with a beneficial outcome. However, young age predicted success. An infrequently occurring adverse event was temporarily increased pain some 6 h post injection.

Conclusion: Anterior rectus sheath blocks using local anesthetics and steroids are safe and long-term successful in more than one-third of children suffering from abdominal pain due to anterior cutaneous nerve entrapment syndrome.

INTRODUCTION

Anterior cutaneous nerve entrapment syndrome (ACNES) is a condition characterized by severe abdominal pain that interferes with daily routines. The incidence of ACNES in a general adult population was estimated up to 55/100 000.¹ Moreover, we recently found that 13% of children with chronic abdominal pain presenting to a pediatrician suffered from ACNES.² The patient's history in ACNES is often nonspecific. Previous surgery, infectious disease, and trauma were identified in half of adult cases.³ One of five children with ACNES had undergone surgery including appendicitis or inguinal hernia repair. It is unclear whether these operations were performed because of the pain, or if the abdominal pain was induced by surgery. Nevertheless, earlier abdominal surgery is possibly a risk factor for the development of ACNES. However, the history of most patients including children is uneventful prior to the onset of ACNES.

The exact pathophysiology of ACNES is unclear but the syndrome is likely caused by events leading to traction or compression of anterior portions of intercostal thoracic nerve endings (thoracic 7–12).^{4,5} A central symptom is a severe localized painful area in anterior portions of the abdomen that is provoked by physical activity.⁶ The small area can readily be identified by thorough examination of the abdominal wall using a palpating finger. Examination of the skin covering the painful area using a swab often reveals sensory alterations such as hyperalgesia, hypoesthesia, allodynia, and/or altered cool perception. Moreover, a positive Carnett's test confirms an origin in the abdominal wall.² Diagnostic tests other than physical examination were hitherto not contributing to the diagnosis but may exclude an underlying visceral disease. Substantial pain relief following administration of a local anesthetic agent probably contributes to the diagnosis.³ The current treatment of choice of ACNES is one (or more) local blocks. If unresponsive, a neurectomy may be considered.^{7,8} However, the efficacy of such a treatment algorithm in children is yet to be determined.

We aimed to study the effectiveness and adverse events of anterior rectus sheath blocks in a prospectively studied series of childhood ACNES.

METHODS

Setting and population

In compliance with local ethical board guidelines for clinical studies, a prospective observational study was conducted at the surgical department of Maxima Medical Center. Maxima Medical Center is a teaching hospital with several areas of expertise including a Center of Excellence for abdominal wall and groin pain syndromes within the Surgery Department.

Study criteria

All patients ≤ 18 years of age who were referred for an alleged abdominal wall pain syndrome between March 2012 and January 2015 were included into the study. Exclusion criteria were absence of ACNES criteria (Table 1), injections under general anesthesia or earlier treatment for ACNES in the referring facility.

Table 1. Criteria for diagnosis of ACNES

1. Presence of a local pain spot in anterior portions of the abdomen
2. Positive Carnett's test
3. Positive pinch test
4. Somatosensory alterations of skin overlying the pain spot (allodynia, hyperalgesia, hypo-/hyperesthesia, and/or altered cool perception)
5. Absence of visceral disease as suggested by history taking, normal routine blood/urine analysis, and/or medical imaging

Data accrual

Each patient who was referred for analysis was standardly requested to complete a questionnaire prior to the first outpatient consultation. This questionnaire determined date of pain onset, pain intensity using a numeric rating scale (PI-NRS, 0 = no pain to 10 = excruciating pain), pain localization, and previous diagnostic and treatment attempts (laboratory tests, medical imaging, medication use, and/or previous interventions). During the first outpatient evaluation, an additional history and physical examination was obtained and registered using a standard protocol. If ACNES was considered likely, the diagnosis was discussed and treatment was initiated.

Anterior rectus sheath blocks

When patients and parents consented to undergo a local block, the skin covering the most painful spot was stretched between two digits of one hand. The other hand holds a 10 ml syringe with a 21 gauge needle was introduced through the skin and the anterior rectus fascia (Figure 1). Needle length matched the thickness of subcutaneous fat. After pulling the plunger ensuring proper positioning (not in a vessel), 5 ml of 1% lidocaine was administered subfascially. Guidance of ultrasound was not used. A block was termed successful if a minimal 50% pain reduction was observed using a pain intensity numeric rating scale (PI-NRS, 0 = no pain to 10 = excruciating pain) ± 15 min later.

Treatment strategy

If this first block was successful, the additional treatment strategy including repeat injections after 2-week intervals were discussed. If the pain had returned at the first 2-week control, a combination of local anesthetic (4 ml lidocaine 1%) and a steroid (1 ml methylprednisolone 40 mg) was administered using the same technique and the effect

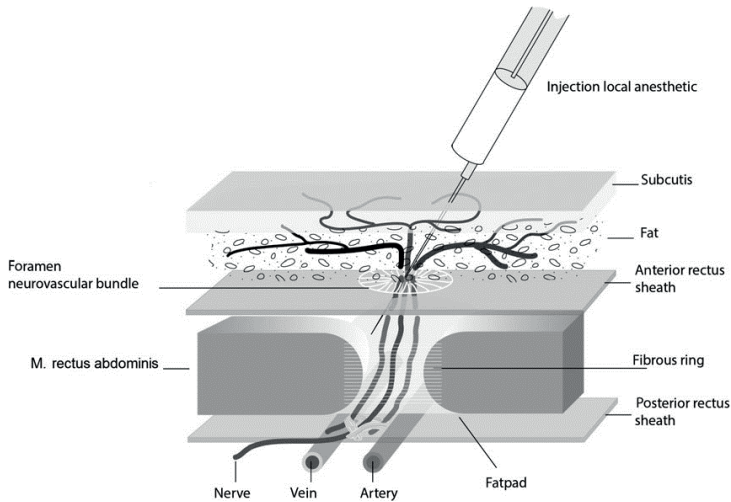


Figure 1. Schematic view of the abdominal wall and the injection site.

was again evaluated after 2 weeks. The total number of blocks was maximized to four. At each follow-up, children were screened on adverse events and the burden of pain. When pain was absent or reduced to an acceptable level not affecting daily routines, patients were dismissed but instructed to contact our facility if the pain recurred. If the effect was consistently short-lived and pain continued to interfere with daily routines after this series of injections, surgery was discussed.

Measures and statistics

Primary outcome measure was the number of patients returning to daily routines without interference of pain following only a regimen of local nerve blocks. Secondary outcomes were occurrence of adverse events or potential relations between various factors (gender, age pain intensity, and duration prior to treatment) and success. Continuous data were expressed as mean [95% CI] or median [minimum-maximum] where appropriate. Comparisons of these data were performed using the Wilcoxon rank test or Mann–Whitney U test and were expressed as mean difference (MD) [95% confidence interval (CI)]. For discrete variables, data were expressed as N (%). The exact test of Fisher was used for their comparison and outcomes were expressed as odds ratio (OR) [95% CI]. A two-tailed P-value <0.05 was considered statistically significant. Statistical analyses were performed using the SPSS 20.0 software (IBM Company, Chicago, IL, USA).

RESULTS

Population characteristics

A total of 91 children were evaluated between March 2012 and January 2015. As six were excluded (all earlier treatment elsewhere), 85 children were analyzed (female, $n = 66$ (78%); age at diagnosis, 15 years (median, range 8–17). A total of 57 children (67%) suffered from a right lower abdomen ACNES. Diagnostic delay was 9 months (median, range 1–48). Pain intensity was severe (median PI-NRS 8, range 6–9), and 76 children (89%) were hindered in sports and school activities. Previous treatments including medication and psychotherapy are listed in Table 2.

Table 2 Treatments prior to ACNES diagnosis ($n = 85$)

Laxatives	24
Analgesics	
NSAID	20
Opioid	10
Paracetamol	7

Only the strongest analgesic is noted if more than one drug is used

First anterior rectus sheath block

After the first abdominal wall injection, PI-NRS decreased from 7.4 [95% CI 7.3–7.7] to 0 [95% CI 0.2–0.7] ($P < 0.01$). A total of 83 children (98%) experienced a minimal 50% drop in pain intensity. At the first follow-up 2 weeks after the injection, 13 children reported persistent pain relief and resumption of daily routines including school, hobbies, and sports (15% initial success rate). The remaining 72 patients experienced a temporary pain relief ranging from several hours to more than a week.

Repetitive blocks

At the first 2 weeks of evaluation, all 72 children with insufficient pain relief opted for a repeat injection. In the following weeks, a regimen of injections (range, 1–3) resulted in prolonged pain relief in 19 children including the 2 not responding to the first injection. At a median 17-month follow-up (range, 4–39 months), a total of 32 children had attained persistent pain relief (38% long-term success rate, Figure 2). Untoward effects possibly associated with the subfascial administration such as hematoma, bleeding, infection, or allergic reactions were not observed. Increased pain levels approximately 6 h after the block were occasionally reported. All remaining 53 children unresponsive to the injection regimen opted for surgery in the following months.

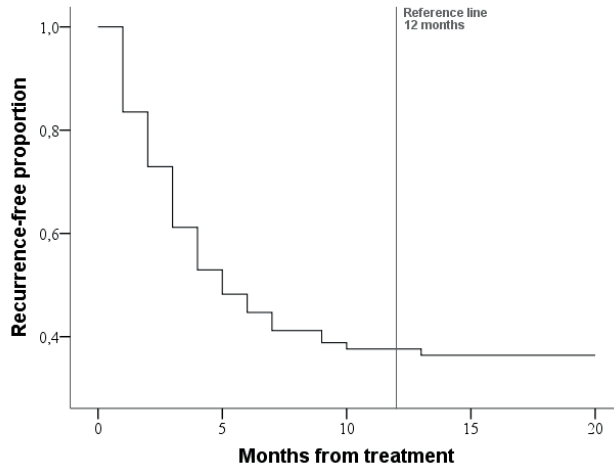


Figure 2. Long-term outcome of anterior rectus fascia blocks in childhood ACNES.

Factors affecting injection outcome

Patients failing injection treatment were significantly older than children reporting success, ($P = 0.04$; mean difference (MD) 1.6 years [95% CI 0.6–2.6]). They also reported a longer diagnostic delay although this difference was not statistically significant ($P = 0.31$; MD 3.7 months [95% CI 2.7–10.1]) (Figure 3). In contrast, treatment outcome was not related to gender ($P = 1$; male OR 0.98 [95% CI 0.43–2.2] vs female OR 1.0 [95% CI 0.8–1.3]). And, also pain intensity was not associated with treatment outcome ($P = 0.18$; MD PI-NRS 0.3 [95% CI -0.1–0.7]).

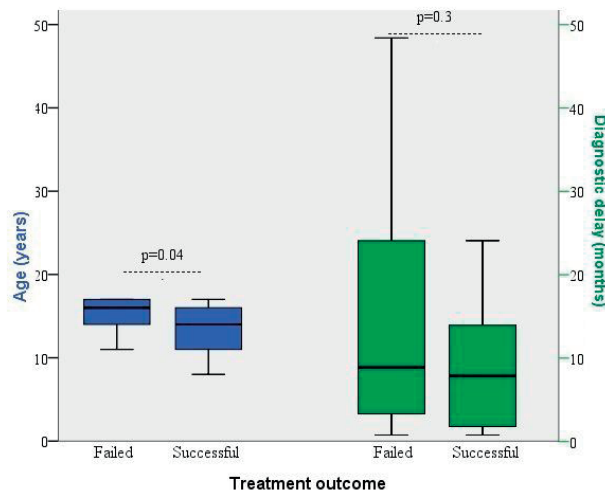


Figure 3. Treatment outcome in relation to age at diagnosis and diagnostic delay in childhood ACNES.

DISCUSSION

In adult ACNES, subfascial injections using a short-acting local anesthetic agent is an accepted first-line treatment once oral pain medication is of no relief. Substantial number of adults respond favorably to this approach and do not require additional treatment such as surgery.³ In contrast, evidence of rectus sheath blocks in pediatric populations is scarce and is limited to a small number of low volume case series. The objective was to prospectively study the effectiveness of such blocks in a large pediatric ACNES population. Analysis of 85 children showed a 38% long-term success rate. Moreover, young age predicted success. The physical and psychological burden of pain associated with ACNES appears tremendous as all remaining 53 children insufficiently responding to blocks chose to undergo surgery.

It is unknown why a subfascial block using lidocaine is often much longer effective than just predicted by the agent's half-life. A similar phenomenon was found in adults with ACNES who experienced permanent pain relief following a regimen of one or more blocks.³ Interestingly, these phenomena were also reported by other groups reporting on childhood ACNES.⁹⁻¹² Lidocaine may influence sodium channels of nerves altering the make-up of various isomers associated with chronic pain.¹³ One may also think that a placebo effect is of influence. However, a randomized controlled trial in an adult population demonstrated superior pain relief following injection of an anesthetic agent when compared to saline.⁵

ACNES patients (of any age) consistently face a substantial diagnostic delay due to the often nonspecific history.¹⁴⁻²⁰ As frequently observed in neuropathic pain disorders, it was assumed that a history of chronic pain was a negative predictor of treatment success. However, success following blocks occurred irrespective of diagnostic delay in the present population. Moreover, intensity of pain was also not related to outcome. These data indicate that any child with any intensity of ACNES may benefit from injections at any time during the course of the disease. Conversely, it may always be worthwhile to administer a rectus sheath block in a child who potentially has ACNES as side effects such as bleeding, infection, or allergy were not observed in the present series.

It was previously proposed that a beneficial response following a subfascial infiltration with lidocaine contributes to diagnosing ACNES.⁵ In the present population, two children did not respond to the initial block but only benefitted after a second trial. An explanation of this finding is lacking but an initial failure may have occurred due to incorrect administration. Therefore, it may be concluded that a beneficial response following a free-hand rectus sheath block is clearly supportive but not conditional of the diagnosis.

The present study harbors flaws including a nonrandomized design. A randomized setting was considered but rejected for ethical reasons. Therefore, it is not possible to

conclude that the effect is merely by injections rather than by a placebo effect. Other limitations are the single-center setting and the use of subjective outcome measures. The use of a free-hand injection technique (instead of ultrasound guidance) may also have influenced treatment success. However, studies comparing the free-hand and ultrasound-guided blocks did not demonstrate either superiority.^{21,22}

In conclusion, a regimen of anterior rectus sheath blocks administered in the outpatient department offered long-term pain relief in more than one-third of childhood ACNES and did not cause adverse events.

REFERENCES

1. Van Assen T, Brouns JAGM, Scheltinga MR et al. Incidence of abdominal pain due to the anterior cutaneous nerve entrapment syndrome in an emergency department. *Scand J Trauma, Resusc Emerg Med* 2015; 23: 19.
2. Siawash M, de Jager-Kievit JW, Ten WT et al. Prevalence of anterior cutaneous nerve entrapment syndrome in a pediatric population with chronic abdominal pain. *J Pediatr Gastroenterol Nutr* 2016; 62: 399–402.
3. Boelens OBA, Scheltinga MR, Houterman S et al. Management of anterior cutaneous nerve entrapment syndrome in a cohort of 139 patients. *Ann Surg* 2011; 254: 1054–1058.
4. Applegate W, Buckwalter N. Microanatomy of the structures contributing to abdominal cutaneous nerve entrapment syndrome. *J Am Board Fam Pract* 1997; 10: 329–332.
5. Boelens OBA, Scheltinga MR, Houterman S et al. Randomized clinical trial of trigger point infiltration with lidocaine to diagnose anterior cutaneous nerve entrapment syndrome. *Br J Surg* 2013; 100: 217–221.
6. Carnett JB. Intercostal neuralgia as a cause of abdominal pain. *Surg Gynecol Obstet* 1926; 42: 625–632.
7. Boelens OB, van Assen T, Houterman S et al. A double-blind, randomized, controlled trial on surgery for chronic abdominal pain due to anterior cutaneous nerve entrapment syndrome. *Ann Surg* 2013; 257: 845–849.
8. Van Assen T, Boelens OB, Van Eerten PV et al. Long-term success rates after an anterior neurectomy in patients with an abdominal cutaneous nerve entrapment syndrome. *Surgery* 2015; 157: 137–143.
9. Skinner AV, Lauder GR. Rectus sheath block: successful use in the chronic pain management of pediatric abdominal wall pain. *Pediatr Anesth* 2007; 17: 1203–1211.
10. Nizamuddin S, Koury K, Lau M et al. Use of targeted transversus abdominus plane blocks in pediatric patients with anterior cutaneous nerve entrapment syndrome. *Pain Physician* 2014; 17: E623–E627.
11. Carnett JB, Bates W. The treatment of intercostal neuralgia of the abdominal wall. *Ann Surg* 1933; 98: 820–829.
12. Bairdain S, Dinakar P, Mooney DP. Anterior cutaneous nerve entrapment syndrome in children. *J Pediatr Surg* 2015; 50: 1177–1179.
13. Akhnikh S, Korte ND, Winter PD. Anterior cutaneous nerve entrapment syndrome (ACNES): the forgotten diagnosis. *Eur J Pediatr* 2014; 173: 445–449.
14. Roumen RM, Scheltinga MR. Abdominal intercostal neuralgia: a forgotten cause of abdominal pain. *Ned Tijdschr Geneesk* 2006; 150: 1909–1915.
15. Applegate WV. Abdominal cutaneous nerve entrapment syndrome (ACNES): a commonly overlooked cause of abdominal pain. *Permanent J* 2002; 6: 20–27.
16. Srinivasan R, Greenbaum DS. Chronic abdominal wall pain: a frequently overlooked problem. *Am J Gastroenterol* 2002; 97: 824–830.
17. Thompson C, Goodman R, Rowe WA. Abdominal wall syndrome: a costly diagnosis of exclusion. *Gastroenterology* 2001; 120(5 Supplement 1): A637.
18. Hershfield N. The abdominal wall. A frequently overlooked source of abdominal pain. *J Clin Gastroenterol* 1992; 14: 199–202.
19. Thomson WHF, Dawes RFH, Carter SSTC. Abdominal wall tenderness: a useful sign in chronic abdominal pain. *Br J Surg* 1991; 78: 223–225.

20. van Assen T, de Jager-Kievit WAJ, Scheltinga MR et al. Chronic abdominal wall pain misdiagnosed as functional abdominal pain. *J Am Board Family Med* 2013; 26: 738–744.
21. Rutten MJ, Maresch BJ, Jager GJ et al. Injection of the subacromial-subdeltoid bursa: blind or ultrasound-guided? *Acta Orthopaedica* 2007; 78: 254–257.
22. Curtiss HM, Finnoff JT, Peck E et al. Accuracy of ultrasound-guided and palpation-guided knee injections by an experienced and less-experienced injector using a superolateral approach: a cadaveric study. *PM & R* 2011; 3: 507–515.