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# Chronic anal pain: A review of causes, diagnosis, and treatment

## ABSTRACT

Chronic anal pain is difficult to diagnose and treat, especially with no obvious anorectal cause apparent on clinical examination. This review identifies 3 main diagnostic categories for chronic anal pain: local causes, functional anorectal pain, and neuropathic pain syndromes. Conditions covered within these categories include proctalgia fugax, levator ani syndrome, pudendal neuralgia, and coccygodynia. The signs, symptoms, relevant diagnostic tests, and main treatments for each condition are reviewed.

## KEY POINTS

Local causes of chronic anal pain can be identified by clinical examination based on index of suspicion and with or without adjunctive diagnostic testing.

Functional anorectal pain syndromes can be subdivided into 3 diagnoses with management individualized for each, albeit with a limited evidence base.

Neuropathic pain syndromes are rare but can be positively diagnosed to allow specific management.

CHRONIC ANAL PAIN IS A RELATIVELY COMMON problem affecting up to 11.6% of the US population.<sup>1</sup> Although many adults have self-limiting symptoms that do not lead to specialist consultation, there is a subgroup of patients with refractory or severe symptoms who do visit surgical clinics. Such patients may see several specialists, such as a colorectal surgeon, urologist, and gynecologist, and may undergo numerous diagnostic or even surgical procedures. It is a sad reality that patients with chronic anal pain commonly feel resigned to defeat when being evaluated by a clinician whose training fails to cover painful anal conditions beyond fissure, fistula, prolapsed hemorrhoids, and other conditions caused by overt disease.

But this need not be so. Clinicians armed with a relatively basic knowledge of possible diagnoses and treatments for chronic anal pain can make a specific diagnosis and initiate treatment even without a complex evaluation.

## ■ DIAGNOSTIC APPROACH AND COMMON PITFALLS

Anal pain can conveniently be grouped into 3 main categories, each with individual diagnoses, causes, and symptoms, which provide a starting point for the examination (Table 1).<sup>1,2</sup> The most common category is local anorectal causes and includes a textbook list of anal conditions that, if persistent, can cause chronic anal pain. These include anal fissure, anal and perineal sepsis (eg, inter-sphincteric fistula or abscess), various ulcerations, and anal tumor.

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**TABLE 1**  
**Main diagnostic categories for chronic anal pain: An overview**

Diagnostic category	Diagnosis or syndrome	Assumed etiology	Main symptoms	Examination findings
Local anorectal conditions	Fissure, perianal sepsis, tumor, ulcers, thrombosed hemorrhoids, severe proctitis	Specific to disorder	Common symptoms: Bleeding, discharge, lump, pruritis ani	Overt findings (may require EUA)
Functional anorectal conditions	Proctalgia fugax	Unknown	Short-lasting (seconds or minutes) sharp deep rectal stabbing or cramping. No radiation. No anorectal pain between episodes	No findings
	Levator ani syndrome	Pelvic floor muscle tension or spasm	Chronic (> 30 minutes) dull rectal ache or pressure sensation. Radiation to buttock, vagina, thigh. Other functional diagnoses common (eg, IBS, FDD, fibromyalgia)	Tender puborectalis, replicates pain (usually left side)
	Unspecified functional anorectal pain	Unknown	Chronic (> 30 minutes) dull rectal ache or pressure sensation. Other functional diagnoses very common (eg, IBS, FDD, fibromyalgia)	No findings
Neuropathic pain syndromes	Coccygodynia	Coccyx trauma leading to peripheral sensitisation	Perineal pain triggered by sitting	Tender on pressure or manipulation of coccyx
	Pudendal neuralgia	Pudendal nerve entrapment: peripherally generated or neuropathic pain	Unilateral perineal pain with paresthesia. Worse on sitting. Nantes criteria <sup>2</sup>	Pain on transvaginal pressure on ischial spine
	Phantom rectum syndrome	Neuropathic pain (deafferentation)	Specific to disorder	Specific to disorder
	Paroxysmal extreme pain disorder	Neuropathic pain (genetic)	Specific to disorder	Specific to disorder

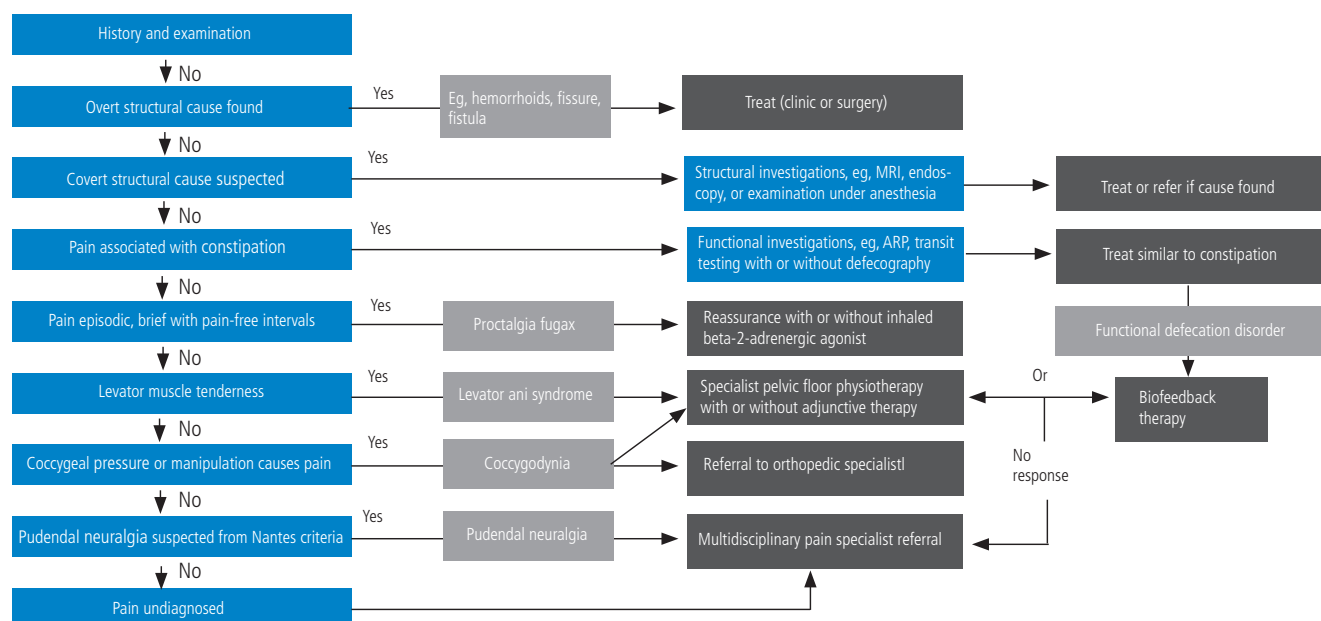
EUA = examination under anesthesia; FDD = functional defecation disorder; IBS = irritable bowel syndrome

Pitfalls to avoid in the diagnosis of chronic anal pain due to local anorectal conditions include the following:

- Attributing the anal pain to hemorrhoids (only thrombosed external hemorrhoids cause significant pain)
- Attributing the pain to a fissure without clear proof of a chronic fissure on examination (under anesthesia, if required), even if this has been “diagnosed” in the past

- Failing to consider less common diagnoses such as ulcers due to Crohn disease, tuberculosis, human immunodeficiency virus, syphilitic chancre, herpes, the vasodilator drug nicorandil (used globally but not approved by the US Food and Drug Administration), proctitis (including pelvic radiation disease), tumor, or solitary rectal ulcer.

These pitfalls may lead to a nonselective approach to diagnosis and to an extensive workup including endoscopy, anorectal physiologic testing, endoanal



**Figure 1.** Algorithm for diagnosis and management of chronic anal pain.

ARP = anorectal physiologic testing; MRI = magnetic resonance imaging

Based on information in reference 3.

ultrasonography, or pelvic magnetic resonance imaging (MRI), with the goal of excluding other diagnoses. While this broad approach with extensive testing can allay anxiety in an anxious patient and possibly set a starting point for treatment, it is costly and may commit the patient to a series of investigations that are invasive, embarrassing, and not cost-effective.

Thus, a selective approach is generally recommended based on suspicion from the patient's history and examination findings of past or present structural disease.<sup>3,4</sup> For example, symptoms of covert perianal sepsis (discharge or swelling as well as pain) or a past history of abscess or anal fistula surgery should prompt MRI even if a fistula is not clinically evident. Similarly, symptoms of obstructed defecation or concomitant fecal incontinence would promote consideration of anorectal physiologic testing and endoanal ultrasonography. **Figure 1** shows an algorithm for the diagnosis and management of chronic anal pain.<sup>3</sup>

### Chronic perineal pain

This review excludes discussion of chronic perineal pain, defined as pain felt between the posterior fourchette (posterior lip of the introitus) and the anus and, in males, between the scrotum and the anus. The diagnosis of perineal pain syndrome requires the occurrence of persistent or recurrent episodic pain that is either related to the micturition cycle or associated with symptoms suggestive of urinary tract or

sexual dysfunction. Although some conditions that cause chronic anal pain can also lead to pain in the perineum, patients meeting the definition of chronic perineal pain should be managed by appropriate specialists (gynecology, urology) to examine for urogenital causes such as episiotomy pain and prostatodynia.

### FUNCTIONAL ANORECTAL PAIN SYNDROMES

If a careful history and digital and rigid endoscopic examination of the anorectum exclude local anorectal conditions, the next most common diagnostic category is functional anorectal pain syndrome.<sup>5</sup> The term functional denotes that structural or biochemical causes are absent on routine evaluation, and it should not be considered pejorative (eg, symptoms are all in the patient's mind). In fact, of the 3 defined syndromes—proctalgia fugax, levator ani syndrome, and unspecified—the first 2 can be positively diagnosed by conducting a careful pain history and examination. The key diagnostic criteria relate to the character and duration of pain and to findings on examination of the levator ani muscle (**Table 1**).<sup>1,2</sup>

#### Proctalgia fugax

This syndrome was described back in 1962 as a condition that is “harmless, unpleasant, and incurable.”<sup>6</sup> Diagnosis is based on a history of sudden-onset pain in the rectal area lasting for only seconds or minutes

**TABLE 2**  
**Randomized, controlled clinical trials of treatments for chronic anal pain**

Author, year	Diagnosis	Intervention	Comparator(s)	Main findings
Eckardt et al 1996 <sup>10</sup> N = 16 (crossover)	Proctalgia fugax	Inhaled salbutamol	Placebo	Salbutamol shortened duration of severe pain vs placebo ( $P = .019$ ); effect most marked in patients having prolonged attacks
Abbott et al 2006 <sup>11</sup> N = 60	Pelvic floor myofascial pain	Botulinum toxin A; pelvic floor injection	Placebo: saline injection	Significant reductions in dyspareunia and pelvic floor pressure with both botulinum toxin and placebo
Dessie et al 2019 <sup>12</sup> N = 59	Myofascia pelvic pain	Botulinum toxin A; pelvic floor injection	Placebo: saline injection	No significant clinical effect
Rao et al 2009 <sup>13</sup> N = 10 <sup>a</sup> (crossover)	Levator ani syndrome	Botulinum toxin A; transanal injection	Placebo	No effect of either botulinum toxin or placebo
Chiarioni et al 2010 <sup>14</sup> N = 157	Levator ani syndrome	Biofeedback	EGS; levator muscle massage	12-month results Pain days: 14.7 (baseline) 3.3 (biofeedback) vs 8.9 (EGS) and 13.3 (massage) Pain intensity: 6.8 (baseline) 1.8 (biofeedback) vs 4.7 (EGS) and 6.0 (massage) Adequate relief: 87% (biofeedback) vs 45% (EGS) and 22% (massage)
Zoorob et al 2015 <sup>15</sup> N = 29	Levator ani syndrome	Steroid injections in levator ani trigger points	Pelvic floor physiotherapy	Both groups improved equally (60% achieved 50% reduction in symptoms)

<sup>a</sup>Only 7 had complete data.

EGS = electrogalvanic stimulation

(mean 15 minutes in 1 study<sup>7</sup>) then disappearing completely.<sup>7,8</sup> The pain can occur night or day and vary in severity from uncomfortable to unbearable.

From a treatment perspective, the problem with diagnosing proctalgia fugax is that symptoms are generally too brief or infrequent to treat. Thus, the key is patient reassurance and explanation, such as describing the condition as a “cramp in your bottom” that is harmless and not indicative of any serious bowel disease. For severe cases, several drugs have been tested including clonidine, nifedipine, diltiazem, nitroglycerine, and even (historically) chloroform.<sup>8,9</sup> However, only inhaled salbutamol (albuterol), a beta-adrenergic agonist, has been investigated in a randomized controlled clinical trial.<sup>9,10</sup> Antidepressants such as amitriptyline or antianxiolytics are sometimes used but have no evidence base as to their efficacy. **Table 2** lists the treatments for chronic anal pain investigated in randomized clinical trials.<sup>10–15</sup>

### Levator ani syndrome

Levator ani syndrome—also called pelvic myalgia, pelvic floor myofascial pain, and pelvic floor muscle spasm—is chronic anal pain resulting from tension or spasms in the levator muscles leading to compression of nerve endings and pain via peripheral sensitization. Patients often describe a dull ache or pressure sensation in the rectum that is exacerbated by prolonged sitting and relieved by standing or lying down.<sup>5</sup> Some patients describe the feeling as like sitting on a ball or having a ball inside their rectum. The pain commonly lasts for hours but may be continuous, with sudden exacerbations.<sup>16,17</sup>

Levator ani syndrome rarely occurs at night. Instead, the pain usually begins in the morning and increases in severity throughout the day. It can radiate into the vagina, the gluteal area, or the thigh. The pain may be precipitated by apparently unrelated factors such as long-distance car travel, stress, sexual

TABLE 3  
Treatments for levator ani syndrome

Category	Examples	Level of Evidence	Comments
Behavior therapy	Biofeedback to improve defecation dynamics	B	Most effective treatment for LAS in single RCT <sup>14</sup>
Muscle relaxant	Electrogalvanic stimulation	B	More effective than massage in single RCT <sup>14</sup> ; benefits decrease in long-term
Muscle relaxant	Diazepam	C	Poorly effective in the long-term; addictive potential
Muscle relaxant	Digital massage of puborectalis muscle	D	No standardized methodology; often provided with sitz bath
Anticholinergic	Botulinum toxin A injection	B	Ineffective as transvaginal or transanal injection in three RCTs <sup>11–13</sup>
Anti-inflammatory	Pelvic floor muscle steroid injection	D	Equally effective as physiotherapy in pilot RCT <sup>15</sup>
Antidepressants	Amitriptyline	D	Unclear mechanism of action; diverse dosage
Neuromodulation	Sacral neuromodulation	D	Conflicting results in small observational studies

LAS = levator ani syndrome; RCT = randomized controlled trial

intercourse, or normal defecation that can potentially lead to stool-withholding.<sup>16,17</sup> Tenderness (reproducing pain) on palpation of the levator muscle (usually the left side, for unknown reasons) is diagnostic.

The overlap of levator ani syndrome with functional defecation disorder<sup>5,16</sup> brings into play several well-established risk factors for the latter that may be determined from the history including anxiety, depression, and a history of sexual abuse.<sup>17–19</sup>

**Treatments.** Of the various treatments that have been studied for levator ani syndrome (Table 3),<sup>11–15</sup> the best evidence is for behavioral training with biofeedback. In a randomized controlled trial of 157 patients, Chiarioni et al<sup>14</sup> compared behavioral training against electrogalvanic therapy (ie, transvaginal or transanal direct neuromuscular stimulation using low-voltage electric charge from a probe) and massage. An intent-to-treat analysis showed that 87% of patients reported adequate relief of rectal pain with biofeedback vs 45% of patients with electrical stimulation and 22% with massage. The improvement was maintained at 12 months.<sup>14</sup>

However, behavioral training with biofeedback is not universally available, and most patients with levator ani syndrome are referred for a comprehensive program of pelvic floor physical therapy focused on pain management. These programs are different from standard pelvic floor physical therapy for pro-

lapse or incontinence that focus on muscle training to strengthen the pelvic floor. Programs for levator ani syndrome include techniques that focus on myofascial release, muscle-stretching, and posture improvement. Most treatment programs are poorly standardized and may include an adjunct such as electrogalvanic stimulation.<sup>20,21</sup> Other attempts at pain management include the Stanford pelvic pain protocol (the Wise-Anderson protocol), which includes relaxation therapy and use of a wand-like device that patients can use to massage internal pelvic myofascial trigger points. The wand was approved by the US Food and Drug Administration in 2012 based on results of a 4-year clinical trial.<sup>22</sup> Local anesthetic injections also have shown efficacy when administered as an adjunct by trained clinicians.<sup>15,23</sup>

**Coexisting and overlapping conditions.** Patients with levator ani syndrome commonly have symptoms of obstructed defecation, and there is a well-acknowledged overlap with functional defecation disorders such as dyssynergic defecation.<sup>5</sup> Biofeedback to improve rectoanal coordination (which includes pelvic floor relaxation) should be the first-line treatment for dyssynergic defecation.<sup>24</sup> Other functional and chronic pain disorders may coexist such as irritable bowel syndrome and fibromyalgia. Attention should be paid to holistic management, especially if depression and anxiety appear to be causing symptoms.

**Botulinum toxin.** If symptoms persist after biofeed-



**TABLE 4**  
**Nantes criteria for pudendal neuralgia by pudendal nerve entrapment**

**Essential criteria**

- Pain in the pudendal nerve area from the anus to the penis or clitoris
- Pain is predominantly experienced while sitting
- Pain does not wake the patient at night
- Pain with no objective sensory impairment
- Pain is relieved by diagnostic pudendal nerve block

**Complementary diagnostic criteria**

- Burning, shooting, stabbing pain, numbness
- Allodynia or hyperalgesia
- Rectal or vaginal foreign body sensation
- Worsening of pain during the day
- Predominantly unilateral pain
- Pain is triggered by defecation
- Presence of exquisite tenderness on palpation of the ischial spine
- Clinical neurophysiology findings in men or nulliparous women

**Exclusion criteria**

- Exclusively coccygeal, gluteal, pubic, or hypogastric pain
- Pruritus
- Exclusively paroxysmal pain
- Imaging abnormalities able to account for the pain

**Associated signs not excluding the diagnosis**

- Buttock pain on sitting
- Referred sciatic pain
- Pain referred to the medial aspect of the thigh
- Suprapubic pain
- Urinary frequency and/or pain on a full bladder
- Pain occurring after ejaculation
- Dyspareunia and/or pain after sexual intercourse
- Erectile dysfunction
- Normal clinical neurophysiology

Reprinted with permission from John Wiley & Sons. From Labat JJ, Riant T, Robert R, Amarenco G, Lefacheur JP, Rigaud J. Diagnostic criteria for pudendal neuralgia by pudendal nerve entrapment (Nantes criteria). *Neurourol Urodyn* 2008; 27(4):306–310. doi:10.1002/nau.20505.<sup>2</sup>

back or pelvic floor physiotherapy, a high dose (total 200 units) of botulinum toxin A (onabotulinumtoxin A) may be injected into the levator (unilaterally or bilaterally). Although the supporting evidence is poor,<sup>11–13,25,26</sup> it is a common practice. It should generally be considered an adjunct to ongoing physical or biofeedback therapy.

## ■ NEUROPATHIC PAIN SYNDROMES

Neuropathic pain syndromes in chronic anal pain are rare compared with local and functional anorectal syndromes. They include coccygodynia and pudendal neuralgia, in which the pain in part has a structural origin, and two overtly neuropathic syndromes, ie,

phantom rectum syndrome and paroxysmal extreme pain disorder (Table 1).

### Coccygodynia

Coccygodynia is pain arising in or around the coccyx depending on its position.<sup>27</sup> The pain is considered to arise from instability of the coccyx with or without pelvic floor spasm.<sup>28</sup> There is usually a history of trauma including childbirth and epidural anesthesia.<sup>29</sup> Risk factors include female sex, obesity, anxiety, depression, and chronic pain elsewhere. Examination will reveal any instability, and movement of the coccyx should reproduce the pain. Rectal examination will often demonstrate coexistent levator ani syndrome.

**Dynamic digital radiography** of the coccyx will show coccygeal instability in about 50% of patients with a clinical diagnosis of coccygodynia.<sup>28</sup> Radiologically, the 2 main patterns of instability are hypermobility (on flexion) and posterior subluxation.

**Management** involves treatment of levator ani syndrome, if present, manipulation of the coccyx, and injection of local anesthetic and steroid into the affected segment.<sup>28</sup> If this fails, an orthopedic referral for coccygectomy may be relevant in selected patients, but this should be done in recognition that outcomes are supported only by retrospective observational data and complications such as infection are common.<sup>29</sup>

### Pudendal neuralgia

Pudendal neuralgia (or pudendal nerve entrapment syndrome) occurs when the pudendal nerve is compressed by the obturator fascia as it forms the Alcock canal.<sup>30</sup> Diagnosis is challenging and requires use of the Nantes criteria, a series of essential, complementary, and exclusion criteria (Table 4).<sup>2</sup> Of these, the essential criteria are most useful as a screening tool. These can be divided into symptom-based and examination-based criteria plus the important confirmatory criterion that pain is relieved by pudendal nerve block. Although this can be accomplished by any trained clinician, it is usual practice to refer the patient to a pain service with neurophysiologic testing expertise so that the pudendal nerve block can be performed under electrophysiologic guidance.

The pain of pudendal neuralgia may be unilateral or bilateral and may radiate to the pelvis and thighs and cause deep pelvic discomfort.<sup>30</sup> A burning sensation and numbness or paresthesia in the gluteal, perineal, and genital areas are commonly reported in association with the pain. Patients with pudendal neuralgia often suffer for several years before being diagnosed.

**Treatments.** Pharmacologic treatments for pudendal

dal neuralgia are primarily tricyclic antidepressants and antiepileptic agents. Simple analgesics are usually ineffective. Pudendal nerve infiltration is another option. It has been shown to have good short-term effects but lacks efficiency in the long-term.<sup>31</sup> Nevertheless, it should always be tried before surgery is contemplated.

Surgical decompression of the pudendal nerve has been proven effective for patients in whom other treatments have failed.<sup>32</sup> Open, laparoscopic, and subgluteal endoscopic approaches for pudendal pain described in the literature include the endoscopic transgluteal minimally invasive technique.<sup>33</sup> Pudendal nerve stimulation using this technique after neurolysis has also shown some success.<sup>34</sup>

## Phantom rectum syndrome and paroxysmal extreme pain disorder

Phantom rectum syndrome (postproctectomy pain) and paroxysmal extreme pain disorder (previously known as familial rectal pain syndrome) are rare causes of chronic anal pain.<sup>35</sup>

Phantom rectum syndrome is a possible diagnosis when an organic source for pain such as perineal hernia or pelvic sepsis is excluded after proctectomy.

Paroxysmal extreme pain disorder is a genetic disorder caused by a mutation in the SCN9A gene. The

patient usually has a family history and onset in the neonatal period or during infancy.<sup>36</sup> It persists throughout life, with autonomic manifestations such as harlequin skin flushing and episodes of syncope with bradycardia. Later in life, the disorder is characterized by attacks of excruciating, deep, burning pain often in the rectal, ocular, or jaw areas. Rectal pain may be triggered by defecation. Management includes use of carbamazepine and needs to be guided by an expert neurologist.

## TAKE-HOME MESSAGES

The key to diagnosis of chronic anal pain is to first exclude specific diseases and then to make a positive diagnosis, which will guide management. It is important to manage patient expectations because outcomes are variable even with a specific diagnosis. For patients with intractable pain despite treatment, referral to a specialist in pain management is recommended. It is important, however, to first clarify the diagnosis and exhaust treatments to avoid the uncertainty caused by parallel or conflicting management strategies. ■

## DISCLOSURES

Dr. Knowles has disclosed board membership, consulting, advisor or review panel participation, and teaching and speaking for Medtronic. Dr. Cohen reports no relevant financial relationships which, in the context of his contributions, could be perceived as a potential conflict of interest.

## REFERENCES

1. Drossman DA, Li Z, Andruzzi E, et al. US householder survey of functional gastrointestinal disorders. Prevalence, sociodemography, and health impact. *Dig Dis Sci* 1993; 38(9):1569–1580. doi:10.1007/BF01303162
2. Labat JJ, Riant T, Robert R, Amarenco G, Lefaucheur JP, Rigaud J. Diagnostic criteria for pudendal neuralgia by pudendal nerve entrapment (Nantes criteria). *Neurourol Urodyn* 2008; 27(4):306–310. doi:10.1002/nau.20505
3. Bharucha AE, Lee TH. Anorectal and pelvic pain. *Mayo Clin Proc* 2016; 91(10):1471–1486. doi:10.1016/j.mayocp.2016.08.011
4. Williams G, Williams A, Tozer P, et al. The treatment of anal fistula: second ACPGBI position statement—2018. *Colorectal Dis* 2018; 20(suppl 3):5–31. doi:10.1111/codi.14054
5. Rao SS, Bharucha AE, Chiarioni G, et al. Functional anorectal disorders. *Gastroenterology* 2016. doi:10.1053/j.gastro.2016.02.009
6. Douthwaite AH. Proctalgia fugax. *Br Med J* 1962; 2(5298):164–165. doi:10.1136/bmj.2.5298.164
7. de Parades V, Etienney I, Bauer P, Taouk M, Atienza P. Proctalgia fugax: demographic and clinical characteristics. What every doctor should know from a prospective study of 54 patients. *Dis Colon Rectum* 2007; 50(6):893–898. doi:10.1007/s10350-006-0754-4
8. Thompson WG, Heaton KW. Proctalgia fugax. *J R Coll Physicians Lond* 1980; 14(4):247–248. PMID:7452532
9. Jeyarajah S, Chow A, Ziprin P, Tilney H, Purkayastha S. Proctalgia fugax, an evidence-based management pathway. *Int J Colorectal Dis* 2010; 25(9):1037–1046. doi:10.1007/s00384-010-0984-8
10. Eckardt VF, Dodt O, Kanzler G, Bernhard G. Treatment of proctalgia fugax with salbutamol inhalation. *Am J Gastroenterol* 1996; 91(4):686–689. PMID:8677929
11. Abbott JA, Jarvis SK, Lyons SD, Thomson A, Vancaille TG. Botulinum toxin type A for chronic pain and pelvic floor spasm in women: a randomized controlled trial. *Obstet Gynecol* 2006; 108(4):915–923. doi:10.1097/01.AOG.0000237100.29870.cc
12. Dessie SG, Von Barga E, Hacker MR, Haviland MJ, Elkadry E. A randomized, double-blind, placebo-controlled trial of onabotulinumtoxin A trigger point injections for myofascial pelvic pain. *Am J Obstet Gynecol* 2019; 221(5):517.e1–517.e9. doi:10.1016/j.ajog.2019.06.044
13. Rao SS, Paulson J, Mata M, Zimmerman B. Clinical trial: effects of botulinum toxin on Levator ani syndrome—a double-blind, placebo-controlled study. *Aliment Pharmacol Ther* 2009; 29(9):985–991. doi:10.1111/j.1365-2036.2009.03964.x
14. Chiarioni G, Nardo A, Vantini I, Romito A, Whitehead WE. Bio-feedback is superior to electrogalvanic stimulation and massage for treatment of levator ani syndrome. *Gastroenterology* 2010; 138(4):1321–1329. doi:10.1053/j.gastro.2009.12.040
15. Zoorob D, South M, Karra M, et al. A pilot randomized trial of levator injections versus physical therapy for treatment of pelvic floor myalgia and sexual pain. *Int Urogynecol J* 2015; 26(6):845–852. doi:10.1007/s00192-014-2606-4
16. Atkin GK, Suliman A, Vaizey CJ. Patient characteristics and treatment outcome in functional anorectal pain. *Dis Colon Rectum* 2011; 54(7):870–875. doi:10.1007/DCR.0b013e318217586f
17. Wald A. Functional anorectal and pelvic pain. *Gastroenterol Clin North Am* 2001; 30(1):243–ix. doi:10.1016/s0889-8553(05)70176-x
18. Leroi AM, Berkelmans I, Denis P, Hémond M, Devroede G. Anismus as a marker of sexual abuse. Consequences of abuse on anorectal motility. *Dig Dis Sci* 1995; 40(7):1411–1416. doi:10.1007/BF02285184

19. Rao SS, Seaton K, Miller MJ, et al. Psychological profiles and quality of life differ between patients with dyssynergia and those with slow transit constipation. *J Psychosom Res* 2007; 63(4):441–449. doi:10.1016/j.jpsychores.2007.05.016
20. Grant SR, Salvati EP, Rubin RJ. Levator syndrome: an analysis of 316 cases. *Dis Colon Rectum* 1975; 18(2):161–163. doi:10.1007/BF02587168
21. Sohn N, Weinstein MA, Robbins RD. The levator syndrome and its treatment with high-voltage electrogalvanic stimulation. *Am J Surg* 1982; 144(5):580–582. doi:10.1016/0002-9610(82)90586-4
22. Anderson R, Wise D, Sawyer T, Nathanson BH. Safety and effectiveness of an internal pelvic myofascial trigger point wand for urologic chronic pelvic pain syndrome. *Clin J Pain* 2011; 27(9):764–768. doi:10.1097/AJP.0b013e31821dbd76
23. Langford CF, Udvari Nagy S, Ghoniem GM. Levator ani trigger point injections: an underutilized treatment for chronic pelvic pain. *Neurourol Urodyn* 2007; 26(1):59–62. doi:10.1002/nau.20393
24. Rao SS, Seaton K, Miller M, et al. Randomized controlled trial of biofeedback, sham feedback, and standard therapy for dyssynergic defecation. *Clin Gastroenterol Hepatol* 2007; 5(3):331–338. doi:10.1016/j.cgh.2006.12.023
25. Nugent E, Beal M, Sun G, Zutshi M. Botulinum toxin A versus electrogalvanic stimulation for levator ani syndrome: is one a more effective therapy? *Tech Coloproctol* 2020; 24(6):545–551. doi:10.1007/s10151-019-02103-w
26. Meister MR, Brubaker A, Sutcliffe S, Lowder JL. Effectiveness of botulinum toxin for treatment of symptomatic pelvic floor myofascial pain in women: a systematic review and meta-analysis. *Female Pelvic Med Reconstr Surg* 2021; 27(1):e152–e160. doi:10.1097/SPV.0000000000000870
27. Traycoff RB, Crayton H, Dodson R. Sacrococcygeal pain syndromes: diagnosis and treatment. *Orthopedics* 1989; 12(10):1373–1377. doi:10.3928/0147-7447-19891001-14
28. Fogel GR, Cunningham PY 3rd, Esses SI. Coccygodynia: evaluation and management. *J Am Acad Orthop Surg* 2004; 12(1):49–54. doi:10.5435/00124635-200401000-00007
29. Karadimas EJ, Trypsiannis G, Giannoudis PV. Surgical treatment of coccygodynia: an analytic review of the literature. *Eur Spine J* 2011; 20(5):698–705. doi:10.1007/s00586-010-1617-1
30. Benson JT, Griffis K. Pudendal neuralgia, a severe pain syndrome. *Am J Obstet Gynecol* 2005; 192(5):1663–1668. doi:10.1016/j.ajog.2005.01.051
31. Tricard T, Munier P, Story F, Lang H, Saussine C. The drug-resistant pudendal neuralgia management: a systematic review. *Neurourol Urodyn* 2019; 38(1):13–21. doi:10.1002/nau.23824
32. Robert R, Labat JJ, Bensignor M, et al. Decompression and transposition of the pudendal nerve in pudendal neuralgia: a randomized controlled trial and long-term evaluation. *Eur Urol* 2005; 47(3):403–408. doi:10.1016/j.eururo.2004.09.003
33. Jottard K, Bruyninx L, Bonnet P, De Wachter S. A minimally invasive, endoscopic transgluteal procedure for pudendal nerve and inferior cluneal nerve neurolysis in case of entrapment: 3- and 6-month results. The ENTRAMI technique for neurolysis. *Int J Colorectal Dis* 2020; 35(2):361–364. doi:10.1007/s00384-019-03480-2
34. Jottard K, Bruyninx L, Bonnet P, Mathieu N, De Wachter S. Pilot study: pudendal neuromodulation combined with pudendal nerve release in case of chronic perineal pain syndrome. The ENTRAMI technique: early results. *Int Urogynecol J* 2021; 32(10):2765–2770. doi:10.1007/s00192-020-04565-1
35. Furtleman CR, Baker MD, Parker KA, et al. SCN9A mutations in paroxysmal extreme pain disorder: allelic variants underlie distinct channel defects and phenotypes. *Neuron* 2006; 52(5):767–774. doi:10.1016/j.neuron.2006.10.006
36. Furtleman CR, Ferrie CD, Aicardi J, et al. Paroxysmal extreme pain disorder (previously familial rectal pain syndrome). *Neurology* 2007; 69(6):586–595. doi:10.1212/01.wnl.0000268065.16865.5f

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