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# Chronic Proctalgia from Leg Length Discrepancy Relieved by a Shoe Insole

Research Article

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#### Abstract

Background: The pathophysiological mechanism behind chronic proctalgia is not fully understood. The study investigated whether treatment of lower limb length discrepancy (anisomelia) with an insole could relieve levator ani syndrome and chronic proctalgia.

Patients and Methods: Nine patients with proctalgia were identified as also having anisomelia. The patients underwent clinical evaluation, including proctoscopy, flexible endoscopy, anal ultrasound and a digital rectal examination to identify tenderness of the levator ani muscle. The patients were treated with an insole worn in the footwear of the short leg, and the effect was observed on proctalgia and tenderness of the levator ani muscle.

Results: The patients had experienced proctalgia for a mean of 41.8 months. The leg length discrepancy ranged from 1-3 cm. Eight patients had left-sided tenderness of the levator ani muscle. The tender levator ani muscle was on the same side as the short leg in five patients and the long leg in four. Two patients showed tenderness on both sides of the levator ani muscle, and three had additional tenderness from behind the rectum to the apex of the coccyx. All patients except one were completely relieved of proctalgia after treatment with a permanent insole worn in the footwear of the short leg, and digital examination showed no more tenderness of the levator ani muscle.

Conclusion: The study demonstrates that chronic proctalgia may be due to the levator ani muscle that has become strained and tender because of pelvic tilting from lower limb length discrepancy, a relationship which until now has been undiscovered. The pain was resolved by wearing a permanent shoe insert to compensate for the short leg. Patients with proctalgia should be examined for leg length discrepancy.

Keywords: Chronic proctalgia; Proctalgia fugax; Levator ani syndrome; Pelvic floor dysfunction; Puborectalis Syndrome; Rectal pain; Anal pain; Anisomelia; Lower limb inequality; Lower limb discrepancy

# Introduction

Chronic or recurrent rectal pain (proctalgia) affects an estimated 6.6% of the population [1]; this condition is only partly understood and is often frustrating for both the patient and the treating clinician. A recent study has provided new insight into the pathophysiology of proctalgia [2]. Among patients reporting tenderness of the levator ani muscles, approximately 85% of subjects exhibited paradoxical contraction or failure to relax the pelvic floor upon straining (dyssynergic defecation) in the absence of symptoms of constipation. Treatment with biofeedback produced a high success rate, superior to that with electrogalvanic stimulation or digital massage.

This paper reports for the first time a relationship between a leg length discrepancy and a tender levator ani muscle, causing chronic proctalgia. The pain was relieved following the use of a permanent shoe insert to compensate for the leg length discrepancy.

# **Patients and Methods**

Between September 2006 and August 2012, nine patients were identified as fulfilling the criteria for having proctalgia, i.e. having recurrent or chronic pain in the anal canal or rectum [3], with alternative disease explanations for these symptoms excluded after multiple diagnostic tests. The patients underwent a standardized work-up, including clinical evaluation, bowel function questionnaire [4], ano-rectoscopy, flexible endoscopy and anal ultrasound. A digital rectal examination was also performed, in which the examining finger was moved from the coccyx posteriorly to the symphysis anteriorly on both sides to explore if tenderness was present in any direction. This manoeuvre often provoked or reinforced the patient's pain. Leg length discrepancy was estimated by measurements from the anterior superior iliac spine to the medial malleolus [5]. A straighten of the angle in the flank on the side of the short leg was noted together with a compensatory flex of the knee of the long leg, with the patient in a standing and resting position.

We then explained to the patients the possible relationship between the anisomelia and the tenderness of the pelvic floor and proctalgia. The treatment was to permanently wear a shoe insert to compensate for the short leg. At follow-up, the patients were questioned about their proctalgia, and the digital rectal examination of the pelvic floor was repeated.

The tenderness of the levator ani muscle was reported as a dichotomous variable, yes (+) or no (-), before and after treatment; other data were expressed as mean and range.

#### Results

The study included four female and five male patients, with a mean age of 60.2 years (range 46-80 years). They had experienced proctalgia for a mean of 41.8 months (range: 3-156 months), which had affected their life considerably. Patient #1 was a male who suffered for over 10 years and had to stop working and running, and periodically required morphine for the pain; he has been treated for haemorrhoids by coagulation. Patient #2 was a male who had been admitted to the hospital several times for intense rectal pain, and a psychological explanation was considered. Patient #3 was an elderly female, who had received a stapled haemorrhoidopexy [6] for haemorrhoids four years earlier; however, her proctalgia first appeared only after she had a leftsided knee alloplasty. Patient #4 was a male who had previously sought treatment from different specialists, and tried physiotherapy and botox injections without achieving relief from the rectal pain. Patient #5 was a female, who, years earlier had a sigmoid resection for diverticulitis and later an operation for cysto-and rectocele. Patient #6 was a male who underwent an operation 12 years earlier for anal fistula, and who experienced worsened proctalgia after longer periods of physical

activity. Patient #7 was a female who had a hysterectomy five years earlier, followed by a colporraphy some years later; she also experienced constipation for many years and has to assist defecation by digital manoeuvres. Patient #8 was a male who also had intermittently use digits to facilitate defecation and who was treated for minor depression. Patient #9 was a female who suffered from constipation for many years, as well as faecal incontinence after childbirth with subsequent surgery; besides proctalgia, she had frequently right-sided lumbar pain extending to the leg.

The measured length of the right legs ranged from 86 to 95 cm and for the left legs 85 to 94 cm. Thus, the individual discrepancies in leg length ranged from 1 to 3 cm (mean: 2 cm). Table 1 lists the specific clinical findings. Eight patients had left-side tenderness of the levator ani muscle. In five patients, the tender levator ani muscle was on the same side as the short leg, and in four patients it was related to the long leg. Patients #7 and #8 showed tenderness on both sides of the levator ani muscle, and three patients had additional tenderness from behind the rectum to the apex of the coccyx.

		Short leg	Long leg	Levator ani tenderness			
				Right		Left	
Pt	Sex			Before	After	Before	After
1	М	L	R	-	-	+	(+)
2	М	L	R	-	-	+*	-
3	F	R	L	-	-	+	-
4	М	R	L	-	-	+*	-
5	F	L	R	-	-	+	-
6	М	L	R	+	-	-	-
7	F	L	R	+*	-	+	-
8	М	L	R	(+)	+	+	(+)
9	F	L	R	-	-	+	-

Table 1: Tenderness of the levator ani muscle before and after treatment of chronic proctalgia with an insole, M: Male, F: Female, R: Right leg, L: Left leg, +: Tenderness of the right or left levator ani muscle, -: No tenderness , (+): Residual tenderness , \*: Tenderness of the anococcygeal raphe posteriorly.

Ano-rectoscopy and sigmoidoscopy revealed internal haemorrhoids in patients #3, #6, and #7. The oldest patient (#3) also exhibited diverticula. Anal sphincter tone and squeeze were normal in all patients, except in patients #4 and #8 (both male), in whom tonus was increased. Anal ultrasound findings were within normal limits in all patients, except in patients #3 and #9 (both female), who exhibited defects in the external sphincters.

The actual bowel function questionnaire showed that the patients had daily defecations with ease, except for patients #2 and #7, who had constipation. None of the patients experienced abdominal pain, but patients #2 and #4 had intermittent bloating. Besides their proctalgia, all patients had shown some colonic co-morbidities [7] and were recommended a high-fibre and low fat diet.

After treatment with an insole for a mean of 5.7 month, the patients were re-examined. The proctalgia had disappeared in all patients, except in patient #8. Digital rectal palpation of the pelvic floor did not elicit pain or tenderness, except in patient #8 and a residual tenderness in patient #1. The patients continued the use of the insole in all footwear.

### Discussion

To the best of our knowledge, this clinical study seems to be the first to demonstrate that chronic proctalgia may be due to pelvic muscles, which have become tender from lower limb length discrepancy. Because of imprecise pain perception, the patient cannot discriminate between pain from the pelvic floor or from the rectum and will feel the pain as coming from the ano-rectal region. Wearing an insole in the shoe of the shorter leg compensated for the limb inequality, leading to relief of the tenderness of the muscles adjacent to the rectum and the proctalgia. This proves indirectly that the leg length discrepancy is the causal factor in these patients.

Patients with rectal pain will usually first be examined for a number of organic diseases, of which the most commonly involved are anal fissure, abscess, haemorrhoids, inflammatory bowel diseases, and cancer; gynaecological, urological and neurological diseases also must be considered [3,8]. However, no organic disease are found in the majority of cases (ca. 85%).

It has been previously assumed that the pathophysiological basis for chronic proctalgia is tension, spasm, or inflammation of the striated muscles of the pelvic floor [9-12]. Psychological assessment of patients with chronic anorectal pain often reveals high anxiety and depression levels [13]. Observations of a psychological pattern in affected patients and their responses to the symptoms have led to proposals of several pathophysiological hypotheses for functional anorectal pain syndromes [8]. Making comparisons with dogs, it has been thought that a spasm occurs from anxiety or stress, leading to the levator ani syndrome [8].

The role of leg length discrepancy as both a biomechanical impediment and a predisposing factor for associated musculoskeletal disorders has been controversial [14]. A retrospective study found that leg length discrepancy of greater than 20 mm affects at least one in every 1000 people [15]. Leg length discrepancy is implicated in a variety of disorders, including low back pain, hip pain and osteoarthritis, sacroiliac malalignment, trochanteric bursitis [14], stress fractures or osteochondritis [16,17], and plantar fascititis [18]. Thus, chronic proctalgia is another possible manifestation of leg length discrepancy. However, it is unknown how often leg length discrepancy occurs without symptoms.

The proctalgia started in our patients when they were aged on line with other researchers [19]. Patients #5, #7 and #9 had undergone pelvic surgery and patient #3 a knee alloplasty. However, all patients had a tender levator ani muscle, which could not be a result of the surgical procedures, except for patient #3. Thus, the leg discrepancy seems to be congenital.

All patients, except one, had tenderness of the left levator ani muscle. In five patients, the tender muscle was on the same side as the short leg. In two patients with a short right leg, additional tenderness was found posteriorly, corresponding to the puborectalis and pubococcygeus muscle and the ano-coccygeal raphe. The mechanism is presumably that a unilateral short leg will cause pelvic tilting and during movement, the body weight will be borne much more by the short limb side [20]. This may lead to chronic strain of the levator ani muscle, resulting in spasm and an inflammatory reaction followed by pain and tenderness, potentially leading to many years of proctalgia. The pain is worsened in the erect position and by physical activity and is relieved when lying down, as experienced by our patients. However, in four patients, the tender levator ani muscle was on the side of the long leg, and one patient experienced additional lumbar, gluteal, and leg pain. It has been reported that some patients react to pelvic tilting by straining the side of the long leg, and a greater amount of pressure is transmitted through the hip of the longer leg [14].

Tenderness of the levator ani muscle is often asymmetric and is found more often on the left than the right side [11,21], which was the case in the present patients. The combination of a short left leg and a long right leg occurred most often in our patients, which is in line with developmental studies of lower limb asymmetries [22] that have found higher incidences of heavier and longer bones on the right side, even in foetuses. The most common treatment for mild leg length discrepancy is the use of shoe lifts, by either wearing a shoe insert or building up the sole of the shoe of the shorter leg; such treatment is highly effective in relieving symptoms of patients with hip pain [23]. In the present patients, the treatment was an insole worn permanently in the footwear of the short leg to equalise the leg lengths and relieve strain on the levator ani muscle, irrespective of the side of the tender levator ani muscle. This treatment led to tenderness and pain relief for all but one patient. Two patients described the result as a small miracle.

Chiarioni et al. [2,3] emphasized the tenderness on palpation of the levator ani muscle as an important clinical and diagnostic sign in levator ani syndrome, but they did not further examine the predominance of left-sided tenderness. They found that 86% of these patients failed to relax pelvic floor muscles when straining to defecate, and 87% were unable to evacuate a water-filled balloon. Furthermore, anal physiological measures revealed that the same mechanism was responsible for both levator ani syndrome and dyssynergic defecation. They also concluded that pelvic floor dyssynergia may present without symptoms of constipation or obstructed defecation. Adequate relief of proctalgia was reported by 87% of patients treated with biofeedback, 45% with electro galvanic stimulation, and 22% with massage. Unfortunately, these authors did not state whether the tenderness of the levator ani muscle disappeared after the treatment.

Another condition involving rectal pain is proctalgia fugax in which pain lasts a few seconds or minutes (described by patients like being stabbed suddenly in the rectum with a spear), with no rectal pain between the episodes. Patients with proctalgia fugax have no tenderness of the pelvic floor muscles during rectal palpation, and the cause of the pain is unclear [19].

In conclusion, a hypothesis may be formulated that a short leg causes pelvic tilting, leading to chronic strain on the levator ani muscle and resulting in pain and palpable tenderness, most often on the side of the short leg. In all patients, except one tenderness of the levator ani muscle and rectal pain disappeared simultaneously by wearing an insole in the footwear of the short limb. Thus, patients with chronic proctalgia should be examined for leg length discrepancy. Future casecontrol studies have to be conducted to clarify the natural history of anorectal pain syndromes, which have to include how often leg length discrepancy may be responsible for chronic proctalgia.

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