

REVIEW

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## Anorectal and perineal pain: new pathophysiological hypothesis

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**Abstract** Anorectal and perineal pain has been described in association with a variety of organic conditions but can also occur under circumstances in which organic disorders are absent and pathophysiology is uncertain. The three most common functional disorders causing anorectal and perineal pain are levator ani syndrome, coccygodynia and proctalgia fugax; Alcock's canal syndrome is also responsible for pain in these areas. We review current concepts about these disorders and the approach to diagnosis and management, and offer a provocative interpretation of the role of psychological factors.

**Key words** Rectal pain • Perineal pain • Levator ani syndrome • Coccygodynia • Proctalgia fugax

### Introduction

Anorectal and pelvic pain may rise from various clinical patterns due to pathologic conditions (gynecological, coloproctologic and urogenital), but there are several circumstances in which the origin is uncertain or the pathophysiological mechanism is not clear: in these cases authors speak about functional anorectal pain syndromes. Clearly this is a pain that is not exacerbated by evacuation. These disorders can be divided into primary and secondary groups according to etiology (Table 1) [1] or defined primarily on the basis of the symptoms (Table 2) [2].

**Table 1** Anorectal pain syndromes

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Primary idiopathic
Secondary
Inflammatory
Pelvic or perirectal abscess
Acute or chronic prostatitis
Inflammatory pelvic disease
Endometriosis
Mechanical
Descending perineum syndrome
Excessive physical activity
Rectal prolapse
Neoplastic
Rectal
Prostatic
Ovarian
Orthopaedic
Coccygeal (trauma)
Alcock's canal syndrome
Neurologic
Multiple sclerosis
Psychiatric
Postoperative
Proctological
Urological
Gynaecological
Orthopaedic

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**Table 2** Classification of syndrome causing anorectal and pelvic pain

Functional gastrointestinal disorders
Anorectal disorders
Functional fecal incontinence
Functional anorectal pain
Levator ani syndrome
Proctalgia fugax
Coccygodynia
Pelvic floor dyssynergia

Functional anorectal and pelvic pain syndromes are characterized by the presence of pain that takes the patient to the attention of the physician. They have been widely described in the literature [1], but their etiology is still under debate. In fact, if their existence is even discussed by some authors who try to explain them with psychosomatic factors, in contrast anatomic and neuroanatomic studies report the possible involvement of functional mechanisms in the explanation of the pain of which a great number of people complain.

Knowledge of the neuroanatomy of the male and female pelvis is a prerequisite to understanding the pathophysiology of anorectal and perineal chronic pain syndromes [3].

### Levator ani syndrome

Also called levator spasm, puborectal syndrome and chronic proctalgia, the levator ani syndrome is characterized by pain described as a dull ache or high pressure sensation in the rectum, exacerbated in the prolonged sitting or supine position, lasting from several hours to some days.

Prevalence of symptoms compatible with this condition in the general population is 6.6% and appears to decline after the age of 45 years; furthermore, it is more common in women [4]. More than 50% of patients are aged between 30 and 60 years.

The origin is most frequently idiopathic, but often related to mechanical factors (e.g. trauma, excessive physical activity in the elderly, compression or straining during uncorrect sitting), psychiatric or postoperative conditions (e.g. anterior resection, radical prostatectomy, hysterectomy and operations on the anal canal such as partial sphincter section, internal lateral sphincterotomy for anal fissure, and internal or external sphincterotomy for transsphincteric fistula). Urinary incontinence after radical prostatectomy leads the patient, spontaneously or with a urologist's advice, to overwork the levator ani in order to overcome the incontinence.

A frequent clinical finding is palpable tenderness of the puborectalis muscle, often with dolorability, as the exam-

ining finger moves from the coccyx posteriorly to the pubis anteriorly. The sign is often asymmetric and is usually found on the left side more than on the right. The diagnosis of levator ani syndrome is made on the basis of the clinical history, rectal examination and exclusion of other diseases that can produce chronic proctalgia.

An international committee proposed diagnostic criteria that consist in chronic or recurrent episodes of rectal pain or aching lasting 20 minutes or longer, for at least 3 months in the absence of other causes such as inflammatory bowel disease, anal cryptitis, intersphincteric abscess, anal fissure, hemorrhoids and coccygodynia [2]. The diagnosis is "highly likely" if symptomatic criteria are satisfied and the physical signs are present; it is "possible" if the criteria are met in absence of physical signs. Furthermore, the pain shows a circadian rhythm: in particular, it is absent during the night, slight in the morning and severe in the afternoon.

The role of anorectal manometry in the diagnosis of levator ani syndrome is not clear: several studies have shown increased anal canal pressure and increased electromyographic activities [5, 6]. One study [5] reported an association between pain relief and a decrease in anal canal pressure. It may be supposed in some cases that a general hypotonic sphincteric apparatus (in multipara women or patients affected by descending perineum syndrome) lead to hypertonia of the puborectalis muscle to balance the minor incontinence, and this hypertonia causes pain. The significance of these observations is uncertain: some authors attributed a predicting role in the evaluation of treatment with biofeedback [6], while others excluded either the diagnostic utility or the predicting response to treatment [7].

The treatment of levator ani syndrome consists in the use of a wide variety of devices, alone or in association, directed at reducing tension in the levator ani muscle. They include techniques such as digital massage, anal dilatation, sitz baths [8], electrogalvanic stimulation, biofeedback [9] and muscle relaxants. Benzodiazepines (e.g. methocarbamol and diazepam) have been reported to be effective for their anxiolytic-sedative and muscle-relaxant effects, due to their agonistic activity on the GABAergic receptors [10]; other muscle relaxants with action on the central nervous system, such as baclofen and cyclobenzepine are also widely used. Local infiltration of botulinum toxin type A, based on the successful treatment of muscular dystonia in torticollis and strabismus, could also be considered among the promising therapies [11, 12]. The literature is rich of anecdotal treatments, but none of them have been evaluated with controlled trials and patient selection criteria varied [1–10].

Surgical treatment [13], consisting of the lateral division of the puborectalis muscle, was first introduced by Kamm et al. [14] and Wasserman [15]. However, this operation should be avoided for the high rate of functional complications, such as major and minor incontinence [16].

Another surgical approach is the partial section of the pudendal nerve or the internal pudendal nerve block; this treatment seems to improve the pain [17].

The functional re-education of the abdominal, dorsal and gluteal muscles seems to affect the stability of the vertebral column and so physical therapy may result in pain relief [18].

Another therapeutic modality is weight loss, since the abdomen and the intra-abdominal viscera weigh on the levator ani muscle and normo- or hypertonia is necessary for continence.

### Coccygodynia

Coccygodynia is pain that arises from the coccyx [19] and is more frequently seen in women and elderly debilitated patients. The pain may be aggravated by sitting on hard surfaces, with poor posture or for long periods, or by prolonged standing, bending or lifting [20].

The majority of cases appear to be in women; the anatomy of the female pelvis leaves the coccyx more prominent than in males and thus more susceptible to injury. The nature of injury and hence the cause of pain may be acute or chronic, or secondary to dislocation or fracture following a fall in the sitting position or subsequent to trauma during vaginal delivery. Chronic trauma may also arise from poor sitting positions or after long car rides and riding tractors and snowmobiles; it may lead to osteoarthritis of the sacrococcygeal and coccygeal joints and result in chronic coccygodynia. It is not clear why

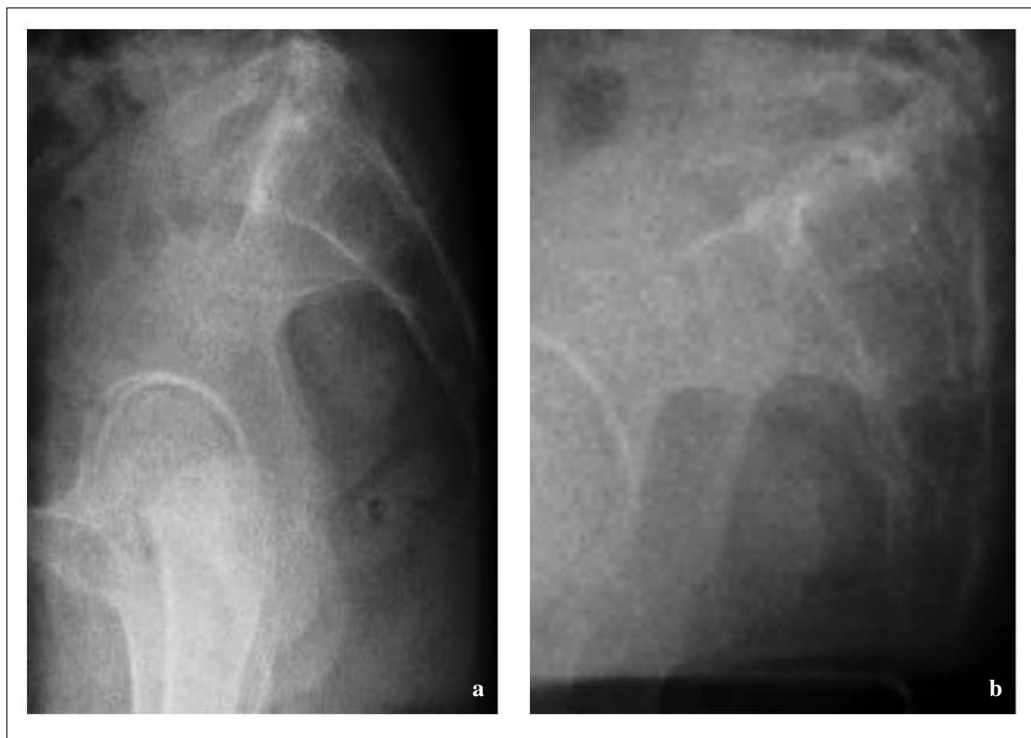
some patients develop a chronic pain syndrome after trauma to the coccyx and others do not. Pain may also be referred to the coccyx from nearby structures (lumbosacral spine, sacrum, anus, rectum, pelvis and genitourinary tract). Other causes of coccygeal pain are anorectal infections and space-occupying lesions, but there are also reported cases of functional coccygodynia [19].

A thorough history should include questions regarding acute trauma to the coccyx following either a fall or birth trauma. A precipitating cause may help the physician in the diagnosis. The patient may describe tenderness in the area of the coccyx, most severe when sitting or more rarely on passing from the sitting to the standing position, but the key to diagnosis is the reproduction of pain when the coccyx is manipulated [10]. Radiographs of coccyx with the patient in standing and sitting positions may support the diagnosis, but not always. In fact Figure 1 shows lateral radiographs of the coccyx in a patient with coccygodynia due to chronic trauma but the radiologic findings are similar to a normal pattern (Fig. 2).

The initial treatment of coccygodynia is conservative and includes protection of the painful coccyx by sitting on a doughnut-shaped pillow to reduce pressure, sitz baths, pelvic relaxation technique and pelvic massage. The first line of management consists of infiltrations of local anesthetic, alone or in combination with steroids. If unsuccessful, Wray et al. [21] introduced the manipulation of the coccyx under anesthesia. Otherwise, in selected patients with severe and unresponsive organic coccygodynia, coccygectomy may be considered.



**Fig. 1a, b** Lateral radiographs of the coccyx in a patient with coccygodynia due to chronic trauma. **a** Standing position. **b** Seated



**Fig. 2a, b** Lateral radiographs of the coccyx in a normal patient. **a** Standing position. **b** Seated

### Proctalgia fugax

The term “proctalgia fugax” suggests a condition characterized by sudden, severe pain in the anal canal or low rectum. The pain lasts a few seconds or minutes (only in about 10% of cases does it last more than five minutes) [14], and then disappears completely leaving the patient asymptomatic until the next crisis. The attack is typically independent from evacuation; it often awakes the sleeping patient or can follow sexual intercourse. Attacks may appear irregularly at any time and at different intervals such as days, weeks, months and even years [3]. In 51% of patients, they occur less than five times per year [2].

The lifetime prevalence of proctalgia fugax ranges from 8% to 18%, and both men and women between the ages of 30 and 50 years are equally affected [22]. Only 17%–20% of affected patients consult a physician.

Unlike levator ani syndrome, patients with proctalgia fugax are asymptomatic during the rectal examination and there are no characteristic physical findings to support the diagnosis, which is based on the clinical description and the exclusion of anorectal disease producing rectal pain, even if palpation of the puborectalis muscle allows the exact localization of the symptom. Harvey stated that at least in some patients proctalgia fugax appears to be an unusual variant of irritable bowel syndrome [23], while Thompson reported that it is common among patients with abdominal symptoms but it is not related to irritable bowel syndrome [24]. Hypertonia of internal anal sphincter has

been reported [7]. A specific autosomal-dominant inherited myopathy of the internal anal sphincter that causes anal pain and constipation in members of two affected families has been described by Kamm et al. [25] and Celik et al. [26], but cannot be assumed to be the cause of most cases of proctalgia fugax.

Patients with proctalgia fugax who sought medical attention were perfectionistic, anxious, or hypochondriacal [27] by psychological testing. The absence of controlled studies makes uncertain the interpretation of these findings, so the role of psychological factors in proctalgia fugax is unclear.

Due to the short duration and sporadic, infrequent nature of the pain attacks, in most cases the explanation of the disorder and the reassurance of the patient are sufficient to guarantee an adequate control of the symptoms. Treatment with calcium antagonists (e.g. nifedipine), clonidine (orally), nitrates associated with a calcium channel blocker such as diltiazem [1], or the beta-adrenergic agonist salbutamol (by inhalation) can shorten the duration of the episodes of pain or suppress the intensity and frequency of anal pain.

### Alcock’s canal syndrome

Alcock’s canal syndrome, or pudendal canal syndrome, is an entrapment syndrome [28] due to injury that may originate from constriction of the internal pudendal nerve in

its musculo-osteo-aponeurotic tunnel between the sacrotuberal and sacrospinal ligaments or during the straddling of the falciform process of the sacrotuberal ligament by the pudendal nerve [29]. It can occur homolaterally or bilaterally with paresthesia or burning pain in the perineal, genital or scrotal areas or in the labia majora, always superficially.

One of the symptoms can be outlet obstruction in case of descending perineum syndrome or incontinence. It affects women more than men, mostly after trauma or prolonged sitting in the elderly.

#### Functional anorectal pain syndrome and psychological aspects

There are several reports of the importance of psychological pattern in patients with chronic idiopathic anorectal pain [7, 27]. The investigation of the psychosomatic components of this condition using standardized and validated questionnaire often reveals abnormally high anxiety and depression levels and personality disorders [30].

Psychological assessment of patients with functional anorectal pain syndrome complements physical examination and clinical investigation in understanding the nature of the condition [30]. In fact a holistic approach, including psychological consultation, may allow a better knowledge of the psychodynamic factors that are the basis of the pain in some patients and may improve the management of these subjects by means of conservative approaches, such as hypnosis, biofeedback, acupuncture and psychotherapy, avoiding further and often ineffective operations [31].

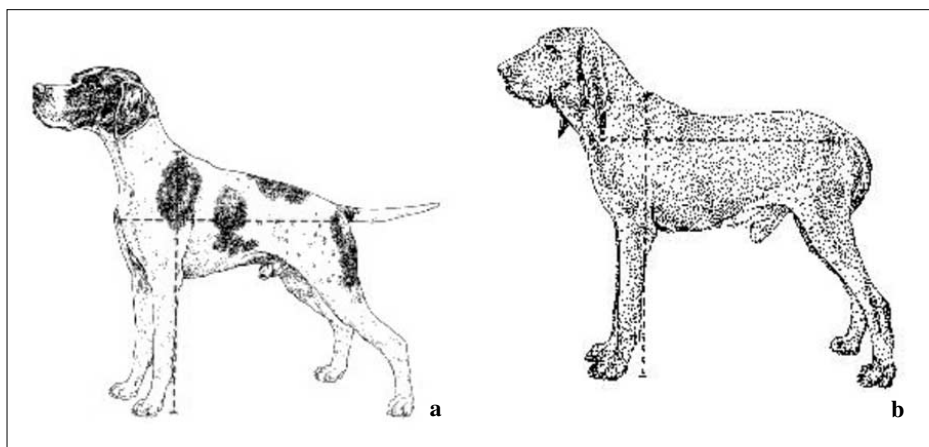
A pathophysiologic hypothesis for the functional anorectal pain syndromes may be proposed on the basis of observations of a psychological pattern in affected patients and their responses to the symptoms. The comparison of man with some animals and their reactions to particular

circumstances of anxiety or stress may suggest a mechanism. For instance, we can observe the position of the dog in various situations (Fig. 3). The English pointer keeps its tail perfectly horizontally while pointing at game. The puborectalis, coccygeus, iliococcygeus and pubococcygeus muscles (Fig. 4a) play an important role in this position. Similarly, a dog reproached by its owner keeps its tail between its legs; this position also requires contraction of the coccygeus, iliococcygeus, pubococcygeus and puborectalis muscles, perhaps to defend the perineum from trauma or sexual abuse.

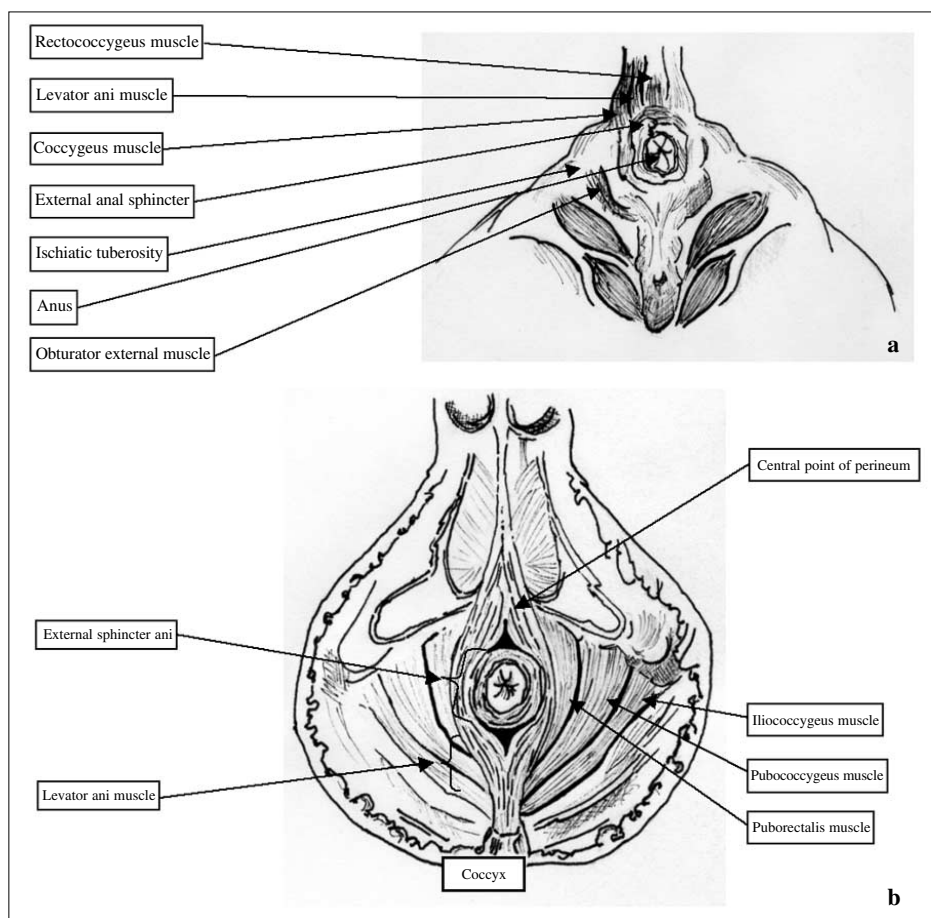
If prolonged for fear or stress, this spasm can also occur in man. The puborectalis muscle (Fig. 4b) moves the coccyx forwards. This could be the cause that leads to the puborectal syndrome (remember that the coccyx is a rudimentary bone of the tail from inferior animals). The coccyx has its own muscular system, deeply reduced in man, represented on both sides by the posterior and anterior sacrococcygeus muscles. The posterior sacrococcygeus muscle moves the coccyx backwards and it is homologous to the extensor muscle of the tail in mammals; in man, this muscle is atrophic. The anterior sacrococcygeus muscle moves the coccyx forwards and is homologous to the flexor muscle of the tail in mammals [32].

Another cause of pain is pressure on the coccyx. In fact, according to the different sitting or supine position, the coccyx may have different angulations. Many patients who suffer from coccygodynia adopt particular positions in order to reduce the pain; for example they often sit on the edge of the chair or mostly on one side (many of them have a sedentary employment, which is particularly stressing). These compressions on the coccyx or slight dislocations are "analgesic positions" that, if abandoned standing up, lead the patient to suffer again.

Finally, a clinical condition similar to what we call levator ani syndrome of man has been observed in some breeds of dogs, in particular in the German shepherd, most frequently in nervous and excitable animals [33].



**Fig. 3a, b** Two different stress conditions of the dog. **a** Pointing to game. **b** Being reproached by the owner



**Fig. 4a, b** Caudal view of the male perineum. **a** Dog. **b** Man

## Conclusions

Functional anorectal pain syndrome is one of the main problems in coloproctology, for both appropriate diagnosis and clinical management. Functional anorectal pain syndromes represent an area worthy of research. The identification of effective treatments for the symptoms requires prospective and randomized trials. We can assume that the pathophysiology is related to a parapsychologic anatomic condition, whose symptoms can be affected by psychological and social factors that, as known, contribute to the pain perception and, furthermore, its cognitive elaboration.

Affected patients should be treated with a multidisciplinary diagnostic and therapeutic approach, often combined with psychological support.

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