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REVIEW

Evaluation and management of chronic anorectal and pelvic pain syndromes: Italian Society of Colorectal Surgery (SICCR) position statement

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Abstract

Chronic pelvic pain is a hidden issue which needs to involve many different usually uncoordinated specialists. For this reason there is a risk that treatments, in the absence of well-defined pathways, common goals, and terminology, may be poorly effective. The aim of the present paper is to summarize the evidence on anorectal pelvic pain, offering useful evidence-based practice parameters for colorectal surgeons' daily activity. Analysis of chronic anorectal and pelvic pain syndromes, the diagnostic and clinical optimal needs for evaluation, and the innumerable low evidence treatments and therapeutic options currently available suggests that a multimodal individualized management of pain may be the most promising approach. The limited availability of dedicated centers still negatively affects the applicability of these principles.

Keywords Anal pain · Perineal pain · Chronic anorectal pelvic pain · Levator ani syndrome · Pelvic pain · Proctalgia · Italian Society Colorectal Surgery · SICCR

Introduction

Chronic pelvic pain is a hidden issue which needs to be managed by many different specialists and which often, in the absence of a well-defined pathway, requires the patient to undergo multiple evaluations that, in the absence of common goals and terminology, ultimately raise the risk of yielding poor effectiveness.

The present work aims to clarify the terminology, clinical approach, diagnosis, and therapies in relation to chronic anorectal pelvic pain (CARPP). Rome III–IV criteria dedicated a brief chapter related to functional rectal pain syndromes, distinguishing proctalgia fugax, levator ani syndrome, and unspecified functional anorectal pain. However, limiting the problem to these pathological entities may be insufficient and all the specialists involved in this field—surgeons, gynecologists, urologists, and physiotherapists—acknowledge the complexity of the medical history of patients with anal pelvic pain and the additional problem that the terminology may vary according to each different specialty involved. Consequently, reliable diagnostic evidence based on well-defined operative algorithms seems to be needed to identify, as fast as possible, the trigger point and the possible causes of the patient's pain.

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As another author has described [1], chronic anal and rectal pain may be classified in three great diagnostic groups: (1) local anorectal conditions—anal fissure, thrombosed hemorrhoids, anal and perianal sepsis, abscess, fistula, ulcerations, severe proctitis (inflammatory bowel disease, radiotherapy), tumor; (2) functional anorectal conditions—levator ani syndrome, proctalgia fugax, unspecified functional anorectal pain; (3) neuropathic pain syndromes like coccygodynia, pudendal neuralgia, phantom rectum syndrome, and paroxysmal extreme pain disorder.

Table 1 summarizes the clinical classification of CARPP. However, it is known that patients with anorectal and pelvic pain very often report a syndrome (with a variety of symptoms) rather than a single painful symptom. Other authors [2] focused on functional pain syndromes and aimed to provide useful diagnostic algorithms to guide subsequent treatment. They classified functional pelvic pain disorders as anorectal (the same classification as above), bladder (e.g., interstitial cystitis/bladder pain syndrome [IC/BPS]), and prostate syndromes (e.g., chronic prostatitis/chronic pelvic pain syndrome [CP/CPPS]).

Rome IV criteria suggest that bowel disorders be considered a continuum rather than independent symptoms [3] and daily experience with patients affected by CARPP confirms this view. The scheme used to analyze the CARPP, with three intersecting circles, is similar to that used by some authors [4], explaining the most frequent symptoms of the pudendal canal syndrome (anal incontinence, perineodynia, urinary incontinence). Undoubtedly, to correctly diagnose patients with CARPP they should be evaluated for all the possible categories (gastrointestinal syndrome, urogynecological/sexual disorders, muscular postural-neuroskeletal, and neurologic disorders) (Fig. 1). Moreover, particular attention is paid to previous surgical procedures: some patients present persistent or chronic (more than 6 months after onset) anal and/or perianal pain after pelvic surgery. Some examples are the stapler procedure, hemorrhoidectomy, fistulectomy, hysterectomy, and prostatectomy. Great care is required to distinguish whether the surgical procedure was the only cause of the pain or just a contributing cause.

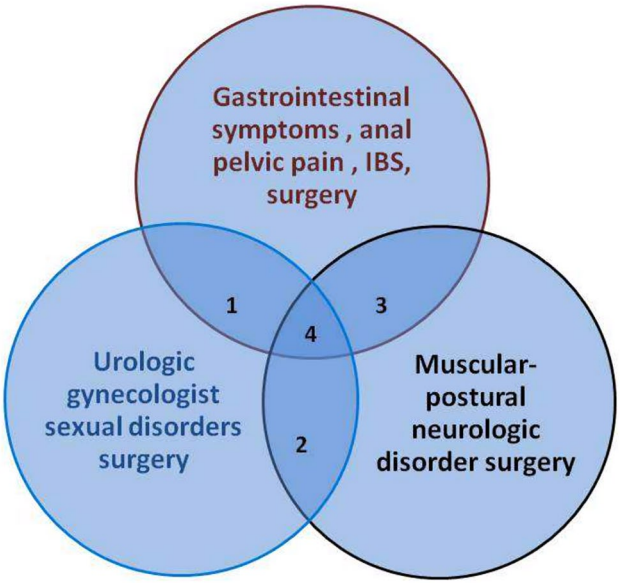

Fig. 1 Multidimensional clinical approach to chronic pelvic pain. The four groups of patients in which multiple triggers and causes coexist may be very complex

The present paper aimed to summarize the evidence on anorectal pelvic pain, offering useful evidence-based practice parameters for colorectal surgeons' daily activity.

Table 1 Main causes of anorectal pain

Etiology	Disorder
Local	Anal (fissure, perianal sepsis, tumor, thrombosed hemorrhoids)
	Rectal (tumor, proctitis, ulcer, prolapse)
	Postsurgery
Functional	Proctalgia fugax
	Levator ani syndrome
	Unspecified functional anorectal pain
Neuropathic	Coccygodynia
	Pudendal neuralgia
	Phantom rectum syndrome
	Paroxysmal pain disorder
Gastroenterological	Inflammatory bowel syndrome
	Ulcerative colitis
	Other colitis/enteritis

Epidemiology and definition

Chronic anorectal pain is estimated to be a common symptom in 11% of the population. However, despite its prevalence and its impact on quality of life, few works have been published in the literature addressing the epidemiology and pathophysiology, making anorectal pain difficult to diagnose and treat [5–8].

Chronic or recurring anorectal pain can be defined as intermittent or constant anorectal pain lasting at least 6 months, in the absence of an underlying anorectal or pelvic disease in the context of a normal clinical examination and investigations [9]. It accounts for a high percentage of proctological consultations and can significantly impact a patient's quality of life, imposing a serious economic and social burden.

Besides the three major disease categories of CARPP (organic, functional, neuropathic), urogynecological

pathologies such as chronic prostatitis, endometriosis, and chronic cystitis also often lead to chronic anorectal pain [10].

Once organic causes of CARPP have been excluded by close patient evaluation and a precise proctological examination as well as instrumental tests, no organic disease will be found in most patients experiencing anorectal chronic pain [6].

Organic anorectal pain

Proctological pathologies may be the cause of chronic pain; these include thrombosed hemorrhoids, chronic anal fissures, anal and perianal suppurations, skin lesions, solitary rectal ulcer, rectoanal prolapse, proctitis, and anal or rectal cancer. Moreover, postsurgical pain (i.e., persistent anal pain after stapled procedures) deserves a separate mention because it may require different treatment strategies [11]. The existence of one of these conditions can be confirmed or eliminated through conventional proctological examination. Sometimes rectoscopy, magnetic resonance, or transanal ultrasound may be necessary.

There are other forms of chronic pain that need to be considered by the proctologist even if they do not have an anorectal origin. Irritable bowel syndrome may be associated with some form of pelvic anorectal pain.

If the clinical history and the physical examination suggest that there is a non-proctological origin of pain, a multidisciplinary approach with input from gastroenterologists, urologists, gynecologists, or pain specialists may be needed.

It is sometimes difficult to collect the patient's medical history when faced with fragile patients with multiple disorders, especially when these conditions are obscured by a pervasive psycho-emotional component. It is extremely important to establish a relationship of trust with the patient in order to plan a proper management program. That is why we need to proceed in stages, focusing on taking a complete medical history, investigating the digestive, genitourinary, sexual, and social spheres. The quality of life should be explored because it may be severely affected.

Functional anorectal pain

When considering functional anorectal pain, the prevalence of levator ani syndrome symptoms in the overall population is 6.6%. More than 50% of those affected are between the ages of 30 and 60 years, and this percentage includes more women than men [5, 7, 12].

Pain is evoked through tension and/or spasm of the pelvic floor muscles that leads to compression of the nerve endings.

For proctalgia fugax the prevalence is difficult to estimate because of the episodic and self-limiting nature of the problem. It ranges from 6% to 18% and is comparable in both men and women [5, 12]. High levels of anxiety disorders, depression, and stress are present in patients suffering from proctalgia fugax and undefined anorectal pain and may act as triggers [12–14].

Patients with functional anorectal pain are often labeled as patients with a psychosomatic disorder and treatments are generally directed at reducing daily stress and promoting muscular relaxation.

According to the Rome criteria, in chronic forms (levator ani syndrome or undefined anorectal pain) the pain lasts at least 30 min with or without tenderness of the levator ani muscle during digital rectal examination. On the other hand, proctalgia fugax is characterized by sudden (usually nocturnal) and recurring anorectal pain which resolves spontaneously within 30 min.

Neuropathic anorectal pain

Coccygodynia, pudendal neuralgia, phantom rectal syndrome, and extreme paroxysmal pain syndrome are the most frequent causes of neuropathic anorectal pain.

The true incidence of coccygodynia has still not been determined. However, factors associated with an increased risk of developing coccygodynia include obesity and rapid weight loss. Women have a five times higher risk of developing coccygodynia than men. External or internal trauma is the most common concomitant factor for coccydynia. External trauma usually occurs from falling backwards, leading to a bruised, dislocated, or fractured coccyx. Minor trauma may occur because of prolonged sitting on hard, narrow, or uncomfortable surfaces. Internal trauma may occur during childbirth, particularly during a difficult labor. Non-traumatic coccydynia includes hypermobility or hypomobility of the sacrococcygeal joint, infection, and variants of coccyx morphology. Neoplasia is an uncommon cause of coccydynia [16–18].

According to estimates by the International Pudendal Neuropathy Foundation, the incidence of this condition is 1 in 100,000 subjects, but the actual prevalence is believed to be considerably higher than reported.

Pudendal nerve entrapment syndrome may affect 1% of the general population and accounts for about 4% of all patient consultations for pain control; women are affected more than twice as frequently as men.

Pudendal neuralgia is typically a "tunnel" syndrome, resulting from cumulative and repetitive microtrauma of the pudendal nerve in the Alcock canal.

The most common causes include repeated pelvic trauma (intensive riding on a bicycle or a horse), prolonged sitting, constipation, stretching trauma due to during childbirth, especially during a difficult delivery, and pelvic surgery. Benign tumors and metastatic lesions to the nerve pathway and herpes simplex infections are infrequent causes of pudendal neuralgia [19–22].

Phantom rectal syndrome and paroxysmal anorectal pain are rare causes of neuropathic anorectal pain. Chronic anorectal pain occurs after abdominoperineal resection in 18% of patients with phantom rectal syndrome, without organic causes such as perineal hernias or pelvic sepsis. The origin of the pain is linked to a nerve deafferentation from loss of afferent inputs in the transmission of nociceptive signals that go from the periphery to the cortex [23, 24]. In chronic paroxysmal anorectal pain, the etiology is traced back to a genetic disease (mutation of the *SCN9A* gene) which encodes the activation of sodium channels involved in the transmission of nociceptive stimuli.

Pathophysiology

Even if various types of pelvic pain-related disorders frequently coexist, several common features can be identified. In particular, the concept of an initial infective or traumatic trigger that, despite having usually resolved, predisposes to the expression and self maintenance of pain.

The initial nociceptive factor induces local secretion of algogenic substances (K^+, H^+, bradykinin, leukotrienes, histamine, substance P). These substances activate the nerve ending of afferent nociceptive fibers and promote the appearance of neurogenic inflammation by antidromic release of substance P and mast cell activation. Intense stimulation of the dorsal horns of the spinal cord by afferent impulses leads to lowering pain thresholds, cellular excitability, and the extension of receptor fields. This extension affects skin, muscle, and viscera innervated by the same metamere, which can explain abdominal wall cutaneous hyperalgesia, myofascial pain, and visceral hyperalgesia. This response beyond the organ itself is characteristic of central sensitization [25]. Alterations of cortical responses to pain in patients with chronic pelvic and perineal pain demonstrated with PET scan and MRI reflect the supraspinal neuroplasticity induced by repetition of the nociceptive message [26].

The painful symptoms vary from one individual to another and according to the individual capacity to activate cortical pain inhibition processes.

Moreover, it must be emphasized that the complex innervation of the pelvic region makes it difficult to obtain adequate identification (and therefore treatment) of the painful area [27].

Another cause of pelvic chronic pain in women is pelvic venous disorders. In this case increased stretch of veins due to obstruction or vasodilation (e.g., in the case of primary ovarian vein insufficiency or iliac vein obstruction and anatomic compression of the left common iliac vein) results in the recruitment of matrix metalloproteinases. These enzymes, if activated, cleave proteins responsible for cell integrity in vein muscle layers and valvular structure. After enzymatic and white cell infiltration secondary to endothelial dysfunction, increased vessel capacitance and worsening valvular incompetence occur. This cyclic mechanism increases venous pressure and develops, in the pelvic viscera, an initial activation of local nociceptors which may result in the clinical presentation of pelvic chronic pain [28]. Potential pathophysiologic mechanisms are summarized in Fig. 2.

Diagnostic evaluation

The clinical examination requires some knowledge of palpatory anatomy and it is often necessary to involve a physical therapist, an osteopath, or a rheumatologist.

The first step is to determine the exact location of pain, asking patients to indicate the most painful site, possibly with the help of visual aids such as pictures or diagrams; this may be decisive in diagnosing pelvic nerve entrapment and pudendal neuralgia (Fig. 3). Accurate investigation of a history of trauma and traumatic or long deliveries should be made.

The neurological examination (sensory, motory, and reflex examination) of the perineum, gluteal region, thoracolumbar segment, and lower limbs should be performed.

In patients with coccygodynia, pain will be much more caudal than in the most common causes of low back pain (located in the upper lumbosacral spine) and more medial than in gluteal pain syndromes (such as obturator internus sacroiliac or pyriform pain).

Hypertonus or spasm of the levator ani muscles is a common finding in patients with anorectal pain, and the function of pelvic floor muscles should be assessed through digital rectal examination and perineal palpation. Anal palpation will assess the sensitivity of the anal sphincter and puborectalis muscle. Furthermore, the location and mobility of the coccyx can be assessed. In addition, the obturator internus muscle, external obturator muscle, piriformis muscle, psoas muscle, ischial spine, ischial tuberosity, pubic symphysis, pubic bone, coccyx, and sacrum should be examined systematically (Table 2).

To objectively quantify the pain, monitor it over time and determine the effectiveness of any medical or surgical treatment, questionnaires can be employed. The most commonly used are Visual Analog Scale for pain (VAS pain), McGill Pain Questionnaire (MPQ), and Chronic Pain Grade Scale (CPGS) [29–31], which consider cognitive and emotional variables.

The validated IBS Severity Scale (IBS-SSS) and the Obstructed Defecation Syndrome (ODS) score could be useful in selected patients when they present with pain of no obvious organic origin to investigate the functional causes, even if they are not specific to objectively quantify the pain.

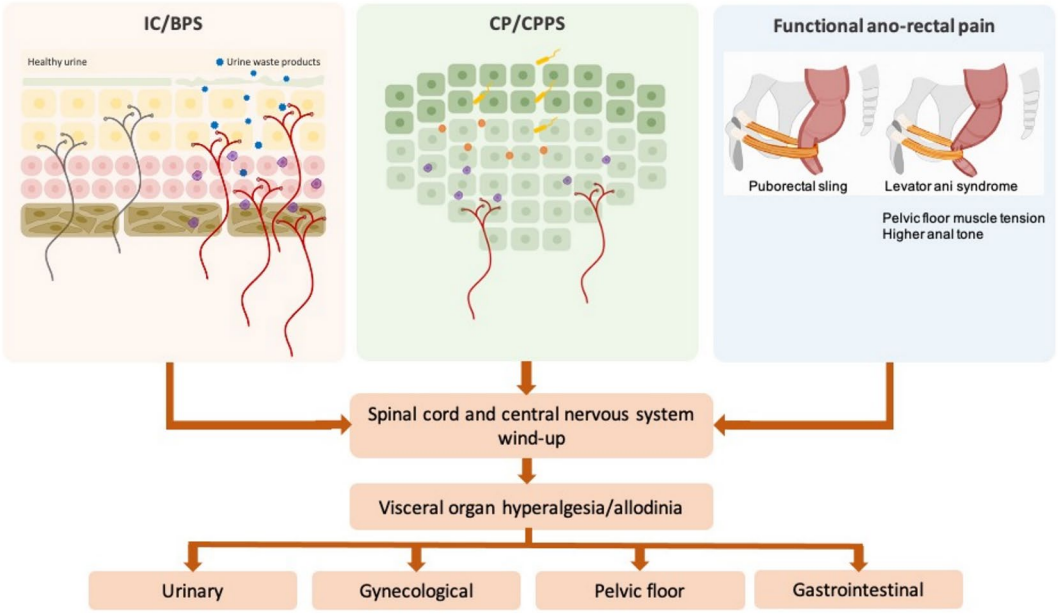

Fig. 2 Pathophysiology of pelvic pain: from left to right the three most common causes of pelvic pain: interstitial cystitis/bladder pain syndrome (IC/BPS), chronic prostatitis/chronic pelvic pain syndrome (CP/CPPS), and functional anorectal pain. In IC/BPS an increase in bladder permeability due to breakdown of the urothelial barrier allows urine waste products (blue) to access the bladder interstitium causing an inflammatory response characterized by immune cell infiltration, mast cell degranulation (violet), that can sensitize and activate nearby afferent nerve endings (red). In CP/CPPS bacterial infection (yellow) may trigger chronic inflammation in the form of autoimmunity directed against prostate antigens with the recruitment of different leukocytes (orange) and mast cells degranulation (violet) causing neural sensitization leading to chronic pelvic pain development. In anorectal pain, functional disorders such as levator ani syndrome and an increased pelvic floor tension play a crucial role in central sensitization

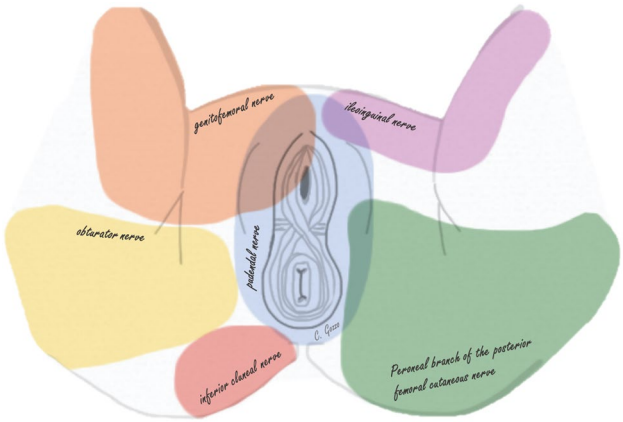

Fig. 3 Innervation of perineum: orange zone, genitofemoral nerve; yellow zone, obturator nerve; red zone, inferior cluneal nerve; green zone, peroneal branch of the posterior femoral cutaneous nerve; purple zone, ileoinguinal nerve; blue zone, pudendal nerve [28, 29]

One of the reasons why CARPP is so difficult to treat is that it is usually multifactorial in etiology, highly heterogeneous in presentation, and there is no test that can be used to confirm the diagnosis with certainty. However, morphological and functional imaging modalities and neurophysiological investigations can play a key role in investigating the organic and the nervous causes of CARPP, especially when the clinical examination and medical history are not sufficient to clarify the etiology. Additionally, they allow us to make a diagnosis of exclusion for all those conditions with an unknown etiology.

Endoanal ultrasound (EAUS) and transperineal ultrasound (TPUS) are used for morphological assessment (thickness, length, and echogenicity) of the internal anal sphincter (IAS), external anal sphincter (EAS), and puborectalis muscle (PR), the integrity of the perineal body, measurement of the anorectal angle, and the dynamic assessment of the

Table 2 Trigger points to be evaluated

Rectal	Transvaginal	Perineal	Gluteal
Puborectalis	Urethral sphincter	Ischial spine	Piriformis
Anal sphincter	Pubococcygeus	Ischial tuberosity	Obturator
Coccyx	Urogenital diaphragm	Pubic symphysis	Cluneal (inferior)
Pubococcygeus		Pubic bone	

posterior compartment. EAUS helps to exclude occult intersphincteric sepsis or occult organic lesions [32]. Additionally, ultrasound abnormalities were found in 22% of patients with functional anorectal pain [33]. The association of a greater PR thickness and paradoxical contraction of PR with chronic proctalgia suggests their potential value as markers of paradoxical contraction of the puborectalis muscle [33, 34].

Anorectal manometry has elicited the possible observation of a dyssynergic muscle contraction and/or hypertonia in the upper part of the anal canal during this syndrome.

Pelvic MRI scan or pelvic CT scan, lower digestive endoscopy, and rectal echo-endoscopy are carried out as part of the positive diagnosis of inflammatory, oncological, or traumatic lesions.

Confirmation of neuropathies requires neurophysiological investigations, which in this case will reveal signs of denervation, mainly of the external sphincter, during electromyography (EMG). It is also possible to find an increase in the latency time of the pudendal nerve, at pudendal nerve terminal latencies (PNTML), when bulbocavernosus or clitoroanal reflexes are studied. Distal lesions of the pudendal nerves were a common feature in women suffering from proctalgia fugax, due to a stretch bilateral pudendal neuropathy [35].

PNTML may be useful in differentiating between radicular or plexus lesions and truncal neuropathy. The latter is associated with a prolongation of the latency time. The fact that electrophysiological abnormalities are unilateral and homolateral to the pain is a strong argument in favor of a positive diagnosis.

Normal cortical somaesthetic evoked potentials rule out a central neurological origin of the chronic pain.

Further investigations (colonoscopy, defecography, etc.) are not systematically useful and should only be considered to confirm or search for an organic lesion origin depending on the clinical context or to evaluate the potential pathological functional alterations involved.

Treatment

Medical treatment

Although the pathophysiology of individual diagnoses is generally multifactorial and poorly understood, growing and widely accepted evidence supports a neuropathic component in chronic pelvic pain syndromes.

Patients with neuropathic pain often present with various combinations of quantitative (hyperesthesia and hypoesthesia), qualitative (e.g., allodynia, dysesthesia, paresthesia), spatial (e.g., faulty localization), and temporal (e.g., after sensation) somatosensory aberrations in the innervation territory of the affected peripheral or central nervous system. Neuropathic pain is challenging to manage, and many patients have pain that is refractory to existing treatments. In randomized clinical trials (RCTs) on pharmacotherapy, no more than half of patients experience clinically meaningful pain relief, and even that is often incomplete. In addition, patients frequently experience burdensome adverse effects leading to discontinuation of the treatment [36]. Despite the innumerable treatments reported, unimodal therapeutic options are mostly unsuccessful, especially in unselected patients. Individualized multimodal management seems to be the most promising approach and may lead to an acceptable situation for a large proportion of patients [37].

NSAIDs

Paracetamol should be considered on its own. It is well tolerated with few side effects. It should be considered an alternative to or given together with nonsteroidal anti-inflammatory drugs (NSAIDs). There is very little evidence for the use of NSAIDs in the management of CPP. Most analgesic studies have investigated dysmenorrhea, in which NSAIDs were found to be superior to placebo and possibly paracetamol [38].

Neuropathic analgesics

Three classes of medications have been recommended as first-line treatments: antidepressants with both norepinephrine and serotonin reuptake inhibition (TCAs and selective serotonin and norepinephrine reuptake inhibitors [SSNRIs]), calcium channel $\alpha_2\delta$ ligands (gabapentin and pregabalin), and topical lidocaine (lidocaine patch 5%). Opioids and tramadol were recommended as a second-line choice, and oxycodone and morphine were considered as third-line treatment [39].

Tricyclic antidepressants Duloxetine and venlafaxine have demonstrated efficacy in RCTs in patients with peripheral neuropathic pain. TCAs have many advantages: low cost, once-daily dosage, and the concomitant effect on depression. The latter is a common comorbidity in patients with neuropathic pain. The major disadvantage of TCAs is the risk of anticholinergic side effects (such as dry mouth, constipation, and urinary retention) and orthostatic hypotension.

Secondary amine TCAs, including nortriptyline and desipramine, are most often recommended because they present fewer side effects and the same benefit on the neuropathic pain component. The treatment should be started at low dosages, administered at night, and titrated slowly, reaching the therapeutic dose in 6–8 weeks.

Anticonvulsants Anticonvulsants have been used in pain management for many years. Although little evidence supports their use in the management of genitourinary pain, they should be considered for possible pain relief.

Gabapentin and pregabalin are medications that bind to voltage-gated calcium channels (at the $\alpha_2\delta$ subunit), producing changes in neurotransmitter release.

It is recommended to start with low dosages and titrate cautiously, to reduce the dose-related dizziness and sedation.

Topical lidocaine When neuropathic pain is well localized, topical lidocaine is appropriate. However, it is unlikely to be efficacious in central neuropathic pain.

Opioid analgesics The use of opioids in urogenital pain is poorly defined. They are not recommended for routine first-line use, primarily because of concerns over long-term safety and side effects (nausea, vomiting, constipation, pruritus, dizziness, dry mouth, sedation). Finally, the risk of opioid misuse, abuse, or addiction in patients with chronic pain cannot be ignored.

Even if opioids are very effective in providing immediate pain relief they require individualized prescriptions because the effective dosage varies considerably among individuals.

Third-line medication These medications should be reserved for patients who do not tolerate or respond to the first- and second-line medications, or for whom the first- and second-line medications are contraindicated [40]. Bupropion, citalopram, paroxetine, and different antiepileptic drugs (e.g., carbamazepine) have been studied in neuropathic pain. Table 3 summarizes the drugs normally used in CARPP.

Non-pharmacological treatment

Pharmacological and surgical therapies are considered the most popular options for treating CPP, but they are not cost-effective and are reported to be related to side effects. Non-pharmacologic therapies have a low risk of side effects and are not contraindicated in patients suffering from drug intolerance or multiple comorbidities in which pharmacological or surgical treatments may be not indicated. This is the reason why more than 50% of patients with CPP are likely to be advised to try non-pharmacological therapies [41].

Psychological therapies

Excluding drug therapies, some psychological [42, 43] treatments have been proposed, mainly in the cognitive behavioral field, for the management of the multiple determinants of the chronic pain experience. The starting point is awareness of one's own pain, and of the psychological, social, and physical reasons determining it. Besides this, improving effective communication with family, friends, and healthcare providers, meditation, mindfulness, enhancing sleep quality, occupational therapy, reducing stress or applying stress management techniques, hypnosis, and group meetings are some of the explored possibilities.

Even if these techniques have been extensively investigated in recent years for many pain syndromes, to find a treatment that may be effective, the literature about pelvic pain is still limited. However, psychological therapies have broad empirical support for their effectiveness, and should be considered in multidisciplinary pain management programs.

Physical therapies

Unlike other chronic pain syndromes elsewhere in the human body, where the musculoskeletal component is often considered secondary to other pathologies, in CPP it is often believed to be prevalent, along with the neurological component, as a primary cause. This is the reason why many physical and rehabilitative interventions such as pelvic floor muscle relaxation, stretch or training, chiropractic care, postural correction, massage and manual therapy, biofeedback, and electromodulation are often adopted.

Table 3 Drugs normally used in CARPP

Drug	Pelvic pain type	Evidence and recommendation
PEA	Neuropathic pain	2C
Paracetamol	Somatic pain	1A
NSAIDs	Chronic non neoplastic	3B
	Dysmenorrhea	1B
Tricyclic antidepressant	Neuropathic pain	1A
Gabapentin/pregabalin	Neuropathic pain	1A
Opioids	Chronic non-neoplastic and neuropathic	1A
Cannabis	Chronic non-neoplastic (endometriosis)	1B

Rather than training muscular activities, tissue lengthening, muscle function restoration, and the ability to improve relaxation are usually required for successful reduction of overactivity symptoms and managing pain in the pelvic region.

Although clinical experience indicates that physical treatments [44, 45] may be effective, evidence-based reports are rare in the literature. Moreover, patients should be informed about potential unpleasant effects in the initial part of the treatment, in which a worsening of symptoms, a prolonged painful sensation, or other changes are possible.

Acupuncture

Acupuncture is a well-known treatment [46] with proven efficacy in musculoskeletal, tumor, postsurgical, chronic prostatitis, and neuropathic, neck, and lumbar pain. Moreover, some studies have reported positive results on pelvic pain, suggesting that acupuncture could be an effective and safe non-pharmacological option for the multidisciplinary management of pelvic pain.

Anesthetic blocks and neuroablation

Neural blockade [47] can be a useful tool both for diagnostic and therapeutic reasons. It should be performed by an experienced operator with an anesthetic background and may involve percutaneously stimulating the peripheral nerves or, under radiological guidance, specific targets such as the superior hypogastric plexus or the ganglion of Walther (ganglion impar).

In many cases, an injection or block will provide only temporary relief and should be repeated. However, in cases of nonpersistent control of pain after multiple treatments, neurolysis or neuroablation can be considered for longer lasting results. There are several different techniques that can be utilized, such as chemo denervation (with alcohol or phenol) or pulsed radiofrequency.

The risk of complications and of inconsistent results due to the multifactorial etiology of pain should always be discussed with the patient before considering this kind of treatment.

Trigger point injections

Trigger point injections may be performed with saline solution, anesthetic, steroids or opioids, isolated or (most frequently) combined or with other treatments. They are recommended for CPP related to myofascial alteration-related pain, nerve entrapment, or muscle spasm.

Even if trigger point injections can provide immediate relief, they may require repeated applications to achieve a full benefit. Moreover, the evidence that injections can be effective regardless of the injectant used suggests a potential placebo effect or a needle insertion effect that has still to be clarified.

Regarding botulinum toxin injection for myofascial pain syndromes the evidence is still inconclusive, and it should be reserved for use in selected patients with overactive bladder with detrusor dysfunction or after the failure of other treatments. As for other treatments, patients should be counseled regarding the potential risks, possible benefits, and expectations before this kind of treatment.

Neurostimulation treatment

Sacral neuromodulation (SNM) appears to be effective for refractory chronic pelvic pain after a successful test phase, yielding pain relief and improving quality of life.

The mechanism of action of neuromodulation appears to be based on the gate control theory and induces pain relief by activating large-diameter Ab fibers while inhibiting smaller Ad and C fibers. Furthermore, neuromodulation could promote the release of endogenous opioids, improve local circulation, and activate the descending inhibitory system. Neuromodulation is a procedure associated with few adverse events, especially when noninvasive transcutaneous electrical nerve stimulation is used. Regarding SNM, to date no RCT has been published. A meta-analysis of 14 studies including 210 patients [48] demonstrated a significant improvement in the VAS pain score ($p < 0.0001$). Furthermore, regarding approaches of SNM implantation, the authors revealed that standard and caudal approaches were both efficacious to reduce pain scores. Finally, a significant improvement in pain was observed both in patients with and without interstitial cystitis/bladder pain syndrome (IC/BPS), although it was lower in the non-IC/BPS group. SNM has proven to be significantly effective in treating voiding symptoms (frequency, urgency, nocturia) associated with IC/BPS. Greig et al. [49], in a recent systematic review and meta-analysis including 26 studies and 853 patients affected by CPP, demonstrated that the implantation rate after test phase success was 64.3%. Significant improvements of pain scores were detected in 13 studies while there was no significant change in three studies. SNM resulted in a mean difference in a 10-point VAS scale of − 4.64. Pain scores improved by 40–53% across studies with full implant patients and the results were maintained after a mean follow-up of 42.5 months (0–59). All studies reported an improvement in quality of life measured by the RAND SF-36 and EQ-5D questionnaires. Complications of Clavien–Dindo grade I–IIIb occurred in 12.5% of patients. In conclusion, SNM seems to be a promising treatment for refractory CP. However, randomized prospective trials are needed to compare SNM with other treatments for CPP.

Percutaneous tibial nerve stimulation may offer pain relief and better quality of life with few adverse events.

Van Balken [50] first introduced the use of percutaneous tibial nerve stimulation (PTNS) in CPP in 2003. Two RCTs have been published regarding the use of PTNS to treat CPP. Gokyildiz et al. [51] compared 12 patients treated with PTNS once a week (in total 12 sessions) with 12 patients in a control group receiving routine intervention. VAS, SF-36, MPQ, and Female Sexual Function Index (FSFI) scores were evaluated. The results showed that women in the experimental group had significant improvements in emotional functioning, mental health, social functioning, and pain as compared to the control group, as well as an increase in FSFI scores.

Istek et al. [52] randomized 33 women with CPP to a PTNS group (16 patients) or control no stimulation group (17 patients). The PTNS group received 30-min sessions for 12 weeks. There was a significant improvement in PPI scores in the PTNS group whereas no change was observed in the control group. Although not statistically significant, there was a slight increase in the PPI-VAS scores in the PTNS group at 6 months of follow-up. There was a significant improvement in all domains of SF-MPQ and SF-36 in the PTNS group that persisted at 6 months of follow-up. No significant change was observed in the control group.

Surgical treatment

Surgical treatment of CPP can be very challenging because of the large number of diseases causing CPP. If urological diseases such as chronic prostatitis, bladder pain syndrome, interstitial cystitis, etc. are the etiological factors of CPP, the patients should undergo surgical evaluation by urologists. Likewise, if a gynecological disease causes chronic pelvic pain, the patient should be referred to gynecologists. The issue becomes more complex when dealing with CPP that must involve a multidisciplinary team, as with endometriosis, vulvodynia, pelvic venous disease, etc. However, here we will consider pathologies for which the role of the colorectal surgeon becomes predominant.

Coccygectomy: this should be reserved for patients with chronic refractory coccydynia unresponsive to conservative management.

The coccygectomy technique, first described in 1937 by Key [53], is a surgical procedure that should be exclusively reserved for patients with coccydynia that is unresponsive to all available conservative therapy and interventional treatment options because of its potential risk of surgical complications such as wound infection, which is reported to be as high as 22% [54]. Although the success rate in coccygectomy ranges from 54% to 100% [55], it is often ineffective when the patient also has lumbar disc disease, and its success rate drops when it is performed in patients with spontaneous idiopathic coccydynia [56].

To date, no RCTs comparing coccygectomy with other conservative or interventional treatments (i.e., stretching/manipulation treatment, local injection of anesthetics and/or corticosteroids, ganglion block, radiofrequency, extracorporeal shockwave therapy) are available. In a single-center retrospective review including 173 patients who underwent coccygectomy for refractory coccydynia, with a mean follow-up of 5.58 ± 3.95 years, there was a significant improvement in average postoperative ODI scores, VAS back pain scores, and many PROMIS29 domains, such as fatigue, sleep disturbance, satisfaction with social role, and PI with daily activities. Using a novel "off-center" wound closure technique, the authors reported postoperative incision site infections in only 16 patients (9.25%), with no differences in long-term outcomes ($p < 0.05$) [57].

In a meta-analysis of 21 studies (18 retrospective and 3 prospective) including 826 patients (75% women) who underwent coccygectomy (720 total and 106 partial) for refractory coccygodynia, the pooled mean difference in pain scores from baseline on a 0–10 scale was 5.03 (95% confidence interval [CI] 4.35–6.86) at 6–12 months follow-up (FU); 5.02 (95% CI 3.47–6.57) at > 12–36 months FU; and 5.41 (95% CI 4.33–6.48) at > 36 months FU. The minimal clinically important difference threshold for pain relief was surpassed at each follow-up. Oswestry Disability Index scores significantly improved postoperatively. The pooled incidence of complications following coccygectomy was 8% (95% CI 5–12), the most frequent of which were surgical site infections and wound dehiscence. The pooled incidence of reoperations was 3% (95% CI 1–5) [58]. Regarding total or partial coccygectomy, there is currently no clarification as to which procedure should be preferred. A prospective observational study on 28 patients who underwent surgical total resection of the coccyx (21 patients) or partial coccygectomy (7 patients) with a mean follow-up of 33 months showed an increased failure rate for pain control associated with partial coccygectomy [59].

In another study, partial coccygectomy was performed in 14 patients and total coccygectomy in eight. The mean follow-up period was 28 months (range 16–48 months). No statistically significant difference was determined between the two groups regarding the mean VAS scores at the final postoperative follow-up examination. The two different surgical methods both had a low complication rate and high patient satisfaction [60].

Surgical release: this is an effective treatment for neuropathic pain syndrome caused by pudendal or cluneal nerve entrapment when other therapeutic measures have failed.

Pudendal and inferior cluneal nerve entrapment syndrome requires a multidisciplinary therapeutic management. When noninvasive multimodal treatments including

pharmacological treatment, physiotherapy, psychotherapy, or nerve blocks fail, surgery can be offered. According to the literature data, surgical nerve release appears to be effective in 60–80% of cases [61]. A positive pudendal nerve infiltration test and fulfillment of all five Nantes criteria seem to be predictive factors of successful surgery [62]. The most common access procedures are by transgluteal and transischiorectal route.

To date, no RCTs comparing surgical pudendal nerve release with other conservative or interventional treatments are available. Furthermore, no RCTs comparing different approaches including open, laparoscopic, or robotic techniques have yet been published. Success in pain resolution appears to be related to complete surgical release of the nerve trunk to allow its total mobility [63].

Jottard et al. [64] described an endoscopic transgluteal minimally invasive (ENTRAMI) approach for pudendal nerve liberation. In a prospective observational trial, 50 patients underwent release surgery using the ENTRAMI technique and were followed up for 1 year after surgery; there was an overall reduction of the average maximal Numeric Pain Rating Scale (NPRS) ($p < 0.05$). Seventy-three percent of patients declared they had a "good treatment response" (patient global impression of change [PGIC] > 30%) and an optimal treatment response (PGIC ≥ 90%) was observed in 40% ($p < 0.05$). No complications were recorded.

In a pilot study, the ENTRAMI technique was used to combine nerve release with pudendal neuromodulation. Sixteen patients were enrolled. At 1 month, the NPRS dropped from 9.5 at baseline to 3.5 ($p = 0.003$), 65% of patients showed a PGIC of > 50%, and an optimal treatment response (PGIC ≥ 90%) was reported in 41% of patients [65].

Multidimensional clinical approach: SICCR practice parameters

When we need to understand the type of patient with CARPP (chronic anorectal pelvic pain) it may be better to use a multifactorial clinical approach to determine which main category they belong to: (a) gastrointestinal symptoms (IBS, stypsis, fecal incontinence); (b) urologic gynecologic disorder (IC, BPS, vulvodynia, sexual disorders); (c) muscular-postural, neuroskeletal, and neurological disorder (spinal disorder, fibromyalgia, orthopedic syndrome). In all these categories, the possibility of previous surgeries should be analyzed: the pain should not always be considered as a complication of a precedent operation, unless there are obvious signs (such as abscess, healing delay, anal stenosis, rigid-stenotic stapler suture line). In addition, it must be asked whether the pain was present before the operation. A patient who needs anal surgery and also has pain with neuropathic characteristics must be scrupulously informed about what it entails, and informed consent obtained. For instance, the surgeon must explain that the surgery may correct the local anatomical/organic disorder but the CARPP might persist or even worsen. For a very complex syndrome like CARPP, if we begin by fitting the patients into categories, solving one group of homogeneous symptoms at a time, we will understand the multifactorial nature of the problem and the need for a multidisciplinary approach. This may also be simple to explain to the patients, creating a relationship of empathy and trust. The multidisciplinary approach also needs specialists for a postural study, like a dedicated physiotherapist or osteopath [66–68], because the muscular component is very frequently a cofactor or an underestimated cause.

The CARPP evaluation diagram is reported in Fig. 4

A possible therapeutic algorithm is summarized in Fig. 5

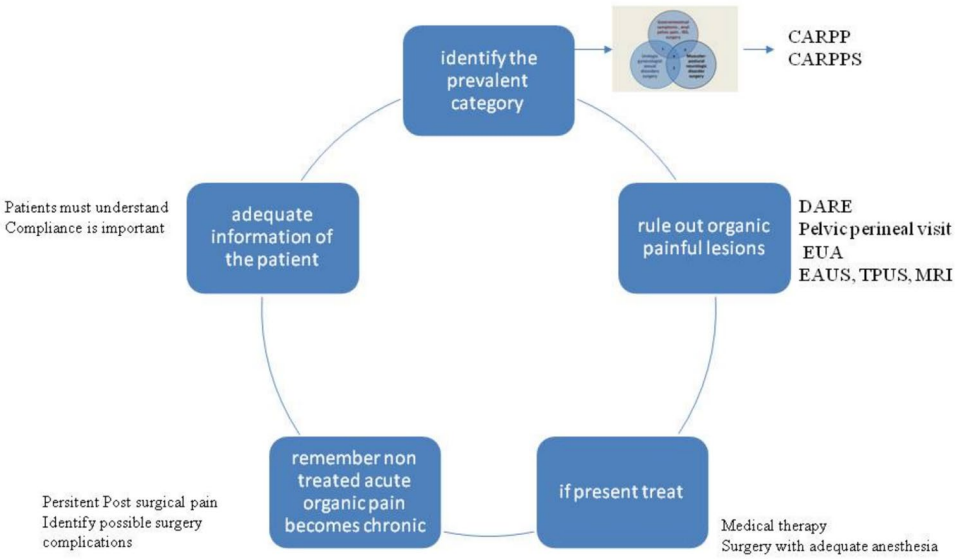

Fig. 4 *CARPP* chronic anorectal pelvic pain, *CARPPS* chronic anorectal pelvic pain secondary, *DARE* digital and rectal exploration, *EUA* evaluation under anesthesia

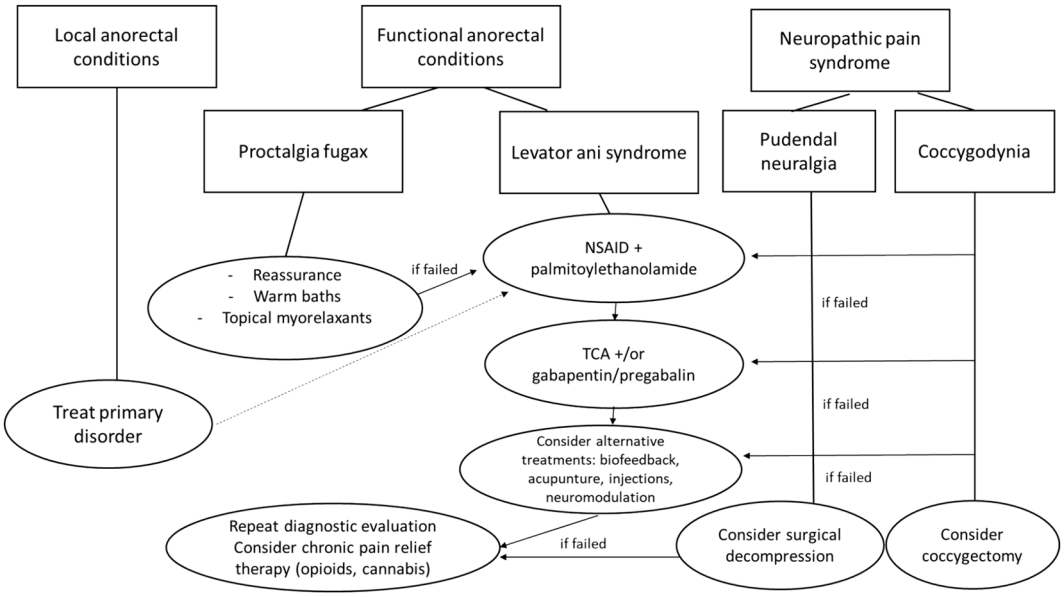

Fig. 5 Treatment algorithm of CARPP

Conclusions

CARPP is a challenging disease to diagnose and treat because of the multiple factors that can cause it and affect its treatment. Significantly heterogeneous terminology is present in the literature but we believe that the denomination CARPP is adequate to identify it. CARPP requires multidisciplinary management that allows the optimization of medical resources and can contain patients' hardships. Finally, analysis of the literature highlights the need to conduct further epidemiological studies to estimate the real-world prevalence of CARPP, as well as prospective randomized studies to optimize its treatment. Our consensus study presents the first attempt to offer, through a review of the literature on CARPP, a guide facilitating shared treatment decision-making in the practice setting.

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Declarations

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