

## Case Report

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# The clinical characteristics of patients with chronic idiopathic anal pain

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**Abstract:** The aim of this study was to investigate the clinical characteristics, treatment outcomes and psychological distress in patients with chronic idiopathic anal pain. The study was conducted on patients referred to Hangzhou Third Hospital for chronic anal pain from January, 2010 to December, 2014. Patient demographics, clinical history, anorectal physiology, and radiological imaging data were recorded for all patients. The treatment outcome was noted for patients treated and followed up for more than 6 month at the present unit. Ninety-six patients with mean age of 45.1 years (range, 17–82) were studied. Seventy-one patients (74.0%) had functional anorectal pain (FARP). The main complaints were dull, sharp, stabbing, or spasm pain. Among all patients, 34.3% reported that their pain radiated into other locations. Fifty-one patients (53.1%) had bowel dysfunction, while 28.1% patients had urinary dysfunction. The common factors associated with pain relief were day time, lying down and warm water baths; the factors that contributed to aggravated pain were night time, defecation or sitting. 92.7% (89/96) of patients reported symptoms of psychological disturbance. FARP patients exhibited increased depression than non-FARP patients ( $P < 0.05$ ). In addition, female patients were more likely to have depression than male patients ( $P < 0.05$ ). The overall pain treatment success rate was 55.2% (53/96). The pain treatment outcome was better in non-FARP patients than in FARP patients ( $\chi^2 = 3.85$ ,  $P < 0.05$ ). Conclusively, chronic idiopathic anal pain is a complex clinical

symptom, involving pelvic floor muscles, the nervous system, endocrine system, and the patients' psychological conditions. Further research is needed to improve diagnosis and treatment for patients with chronic idiopathic anal pain.

**Keywords:** Chronic anal pain; Clinic characteristics; Psychological distress; FARP

## 1 Introduction

Chronic idiopathic anal pain is not a devastating disease defined as a recurrent or persistent pain in the anal canal that lasts more than 3 months without lesions of the anus and rectum, such as anal fissure, complicated hemorrhoids, and perianal suppurative conditions [1-3]. Although anal and perianal pain are common symptoms in colorectal practice, patients with chronic anal and perianal pain are rare [3-5]. However, the prevalence of patients with chronic idiopathic anal pain has increased in recent years. The pain is usually severe, deep, and long-lasting, which constitute a significant constant threat to patients' life quality [6]. Although new diagnostic techniques such as pudendal nerve terminal motor latency measurement, sacral nerve stimulation, botox injection, biofeedback and transanal ultrasound have been made available [1, 7-9], the unknown etiology leads to poor diagnosis and treatment outcomes in patients with chronic anal pain and generally causes psychological disorders such as emotional disorders, depression and anxiety [4, 10-13]. Therefore, the study of psychological factors and clinic characteristics that are implicated in the incidence of chronic anal pain and associated treatment outcomes may be of assistance in clinical practice.

The purpose of the present study was therefore to examine the demographic and clinical characteristics, and associated psychological comorbidities to gain a better understanding for chronic idiopathic pain. A com-

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parative study of the common pain treatments and management procedures among patients in our clinic, and their effectivity was similarly performed.

## 2 Methods

### 2.1 Patients

This study was carried out on outpatients that were referred to the Colorectal Unit of the Hangzhou Third Hospital for chronic anal pain between January 2010 and December 2014. The diagnosis was based on anal finger examination, perianal ultrasound, colonoscopy and/or pelvic MRI that were performed on these patients. Patients with anal fissure, anal fistula, complicated hemorrhoids, perianal abscess and tumor were excluded. The patients with associated different organic diseases (including known spinal lesions, primary disease of nervous system, etc), were not included in the study. The 96 eligible patients had pain in the anal region for more than 3 months with no other local abnormalities found at initial clinical evaluation, were able to read and write, and gave written informed consent prior to enrolment. The study was approved by the Medical Ethical Board of Hangzhou Third Hospital.

### 2.2 Demographic and clinical measures

After obtaining the patients' written informed consent, participants were instructed to fill out a medical history questionnaire including demographic information.

### 2.3 Pain measure

All subjects filled out a 10cm Visual Analogue Scale (VAS) to evaluate the intensity of pain. VAS ranged from 0 ("absence of symptom") to 10 ("maximum intensity of symptoms").

### 2.4 Psychological assessment

Psychometric evaluation was focused on anxiety and depression. The level of depression was assessed by the Beck Depression Inventory (BDI-II) [11, 13, 14] containing 21 multiple-choice items, each with a score from zero to three, so that the total point score varies from 0 to 63. Each item coded from 0 to 3 constituted the basis for a specific

4-level Likert scale used to characterize the psychological profile of the patient. According to the score, the patients were grouped into two categories: high depression (15–63) and low depression (0–14) [15].

Anxiety was assessed by the State and Trait Anxiety Inventory, composed of two axes (A1 for state anxiety and A2 for trait anxiety). Both A1 and A2 are consisted of 20 multiple-choice items; each item has a score from one to four, so that the total point score of A1 and A2 axes can range from 20 to 80. In the general population the average value for the State-Trait Anxiety Inventory (STAI) is  $50 \pm 10$ . This test was selected for its simplicity, validity, and reliability [16, 17].

### 2.5 Bowel function measurement

Information regarding the patient's bowel function was collected using the Wexner constipation score questionnaire and FISII incontinence questionnaire [18, 19].

### 2.6 Anal pain diagnostic criteria

Functional anorectal pain (FARP) was diagnosed using the Rome III diagnostic criteria for functional bowel disorders [20]. A Chronic Proctalgia was diagnosed if patients exhibited chronic or recurrent rectal pain or episodic aching each lasting 20 minutes or longer for at least 6 months, with no other causes of rectal pain such as ischemia, abscess, fissure, hemorrhoids, and coccygodynia. Patients with Chronic Proctalgia were further categorized into Levator Ani Syndrome (tenderness during posterior traction on puborectalis) and Unspecified Functional Anorectal Pain (no tenderness during posterior traction on puborectalis) based on digital rectal examination. Proctalgia Fugax was diagnosed if patients had recurrent episodes of pain localized at the anus or lower rectum, with pain episodes lasting from seconds to minutes and there is no anorectal pain between episodes.

### 2.7 Grouping

Since many patients in this study could be diagnosed as FARP, the patients were divided into two groups, FARP and Non- FARP for better comparison and analysis. The patients in the Non- FARP group did not meet the diagnostic criteria for FARP.

## 2.8 Treatment

After a definite diagnosis, patients received biofeedback and other symptomatic treatments, and those with chronic prostatitis, chronic pelvic pain Syndrome (CP-CPPS), interstitial cystitis, painful bladder syndrome (IC-PBS) received further treatment in the Department of Urology. All kinds of hearing or visual biofeedback (e.g. balloon sensory training, manometry biofeedback, EMG biofeedback) were taken into consideration. Biofeedback therapies were done by a healthcare expert in our institution.

## 2.9 Follow up

The patients were followed up for more than 6 months. The pain treatment outcomes were recorded every three months. The scoring was performed by a single investigator to minimize bias. Treatment success was graded as 0 (no improvement), 1 (temporary improvement), 2 (slight improvement), 3 (marked improvement), and 4 (pain free).

## 2.10 Statistical analysis

Data were analyzed by t-test or  $\chi^2$ -test (SPSS17.0). For all tests, P values less than 0.05 were regarded as statistically different.

# 3 Results

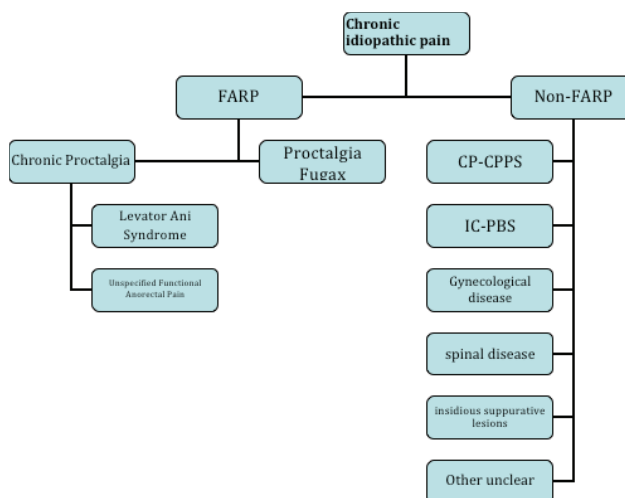
## 3.1 Demographics

96 patients composed of 62 females and 34 males with mean age of  $45.1 \pm 13.5$  years were included in our research. The median of symptoms duration at presentation was 32 months (range, 3–564). Seventy-one patients (74.0%) had functional anorectal pain (FARP) including 22 patients with Levator Ani Syndrome, 37 patients with Unspecified Functional Anorectal Pain, and 12 patients with Proctalgia Fugax. Among 25 non-FARP patients, 6 had CP-CPPS, 8 had IC-PBS, 4 patients had gynecological pelvic inflammatory disease, 2 had spinal disease, 3 had insidious suppurative lesions, and 2 patients had an unclear diagnosis (Figure 1). There were more female patients in the FARP group than the non-FARP group (50/71, 70.4% vs 12/25 48%;  $\chi^2=4.06$ ,  $P<0.05$ ). There was no difference in age

between the FARP group and the non-FARP group (mean age, 43.4 vs 47.2 y;  $P=0.084$ ).

## 3.2 Clinical history

34.3% of patients reported that their pain radiated into other locations such as vagina, perineum, thigh and the lower back. The number of patients with radiating pain was higher in the non-FARP group compared to the FARP group (19/71 26.8% vs 14/25 56%;  $\chi^2=7.01$ ,  $P<0.05$ ). Of all patients, 53.1% (51/96) reported continuous pain, whereas 46.9% (45/96) reported intermittent pain. Patient had mainly dull sharp, stabbing or spasm pains while a few number of patients had burning pain (Table 1).



**Figure 1:** The classification of chronic idiopathic anal pain in this study

**Table 1:** Details of the anal pain.

	All patients (N=96)	FARP (N=71)	Non-FARP (N=25)
Types of the pain			
Radiated pain	33	19	14
intermittent pain	57	42	15
continuous pain	39	29	10
character of the pain			
dull	39	30	9
sharp	21	18	3
stabbing	19	14	5
spasm	12	10	2
burning	14	11	3

51 patients (53.1%) had symptoms similar to those of bowel dysfunction. The most common symptom of bowel dysfunction was obstructed defecation. Fecal incontinence was found in 10.5% of patients while 6 patients reported anal tenesmus. 27 patients (28.1%) had complications associated with urinary disorders. The occurrence of urinary symptoms in patients was greater in the Non-FARP group than the FARP group. (11/71 7 0.4% vs 16/25 48%;  $\chi^2=21.52$ ,  $P<0.05$ ) (Table 2). Overall, 33.3 % (32/96) of patients reported a precipitating event indicating the onset of their symptoms (Table 3).

Also, 30.2 % (29/96) of patients reported factors that relieved the pain (Table 4), the most common being day time, lying down, and a warm water bath. 42.7% (41/96) of patients reported factors that aggravated the pain,

the most common being night time, defecation or sitting (Table 4).

### 3.3 Psychological profile

92.7% (89/96) of patients with chronic anal pain reported symptoms of psychological disturbance including depression and anxiety. FARP patients exhibited greater depression compared to the non-FARP patients ( $p < 0.05$ ). Female patients had a higher mean depression score than male patients ( $p < 0.05$ ). While no difference in depression score was observed between men and women among the non-FARP group, women had significantly higher levels of depression compared to men in the FARP group. There was no significant difference in anxiety scores among the different groups (Table 5 and 6).

**Table 2:** Symptoms of bowel and urinary dysfunction

	All patients (N=96)	FARP (N=71)	Non-FARP (N=25)
Bowel	51	43	8
Constipation	41	36	5
Incontinence	10	9	1
Anal tenesmus	6	4	2
Urinary	27	11	16
Frequent Micturition	13	4	9
Urinary incontinence	4	3	1
Micturition endless	10	4	6

**Table 3:** Initial event marking the onset of the anal pain

	All patients (N=96)	FARP (N=71)	Non-FARP (N=25)
None	64	46	18
Childbirth	5	4	1
anal surgery	6	5	1
Pelvic surgery	2	2	0
Physical activity	3	2	1
Straining stool	4	4	0
Gastroenteritis	5	3	2
Anal sex	2	1	1
Anal trauma	2	1	1
Anal fissure	3	3	0

**Table 4:** The common pain relieving and aggravating factors

	All patients (N=96)	FARP (N=71)	Non-FARP (N=25)
Relieving factors	29	21	8
None	67	50	17
Lying down/relaxation	6	4	2
Physical activity	2	1	1
Digital anal	4	3	1
Warm water bath	7	5	2
Passing flatus	2	2	0
Day time	8	6	2
Aggravating factors	41	32	9
None	55	39	16
Defecation	9	8	1
Sitting	6	5	1
Emotional stress	4	3	1
Physical activity	3	3	0
Sex/orgasm	3	2	1
Lying down/relaxation	2	1	1
Micturition	4	2	2
Night time	10	8	2

### 3.4 Treatment outcomes

Our results showed that the pain management outcome for patients with chronic anal pain was far from ideal. Among all patients that received biofeedback treatment, only 15 were completely healed, 38 patients had their pain alleviated, while no change in pain before and after treatment was recorded for 41 patients. In addition, 2 patients had their pain worsened following treatment. The treatment efficiency was 55.2% (53/96). The pain treatment outcomes were better in the non-FARP group than in the FARP group (35/71 49.3% vs 18/25 72%;  $\chi^2=3.85$ ,  $P<0.05$ ) (Table 7).

## 4 Discussion

The pathophysiology for chronic anal pain is complicated and might be accompanied with some hidden diseases. For example, in our study we found that most patients with chronic anal pain had FARP or disorders such as CP-CPPS, IC-PBS. In addition, we recorded two patients with chronic anal pain caused by spinal abnormalities. One patient had a small spinal tumor, and another had problems with the lumbar intervertebral disc. We had three patients with

small infections located in deep sites that were hard to identify; we were only able to locate these foci of infection with MRI examinations. This suggests that MRI is efficient for the diagnosis of some types of chronic anal pain. This was corroborated by previous studies stating that MRI is helpful for the diagnosis of chronic anal and perianal pain [21]. However, some patients can have chronic anal pain without identifiable abnormalities even with MRI assistance, which leads to difficulties in diagnosis.

The anus is located at the end of the digestive tract and in close proximity to the urinary tract and reproductive organs. The anus is also surrounded by peripheral nerve fibers and in a close relationship to the endocrine system. As a result, disorders in any of these related or adjacent systems may lead to chronic anal pain [22]. For example, some of our CR-CPPS and IC-PBS patients had pain mainly located in the anus region. Women with gynecological inflammation may also experience pain in the anus.

Patients with chronic anal pain often experience urinary or defecation dysfunctions. In the present report, 51 patients experienced constipation and 10 patients exhibited fecal incontinence. This was supported by findings that constipated patients frequently experience anal pain [23, 24].

**Table 5:** Depression and anxiety scores for FARP and non-FARP patients

	All patients (N=96)	FARP (N=71)	Non-FARP (N=25)
Depression	13.21±0.68	14.20±0.88	10.40±0.56
State Anxiety	44.80±1.52	45.07±1.87	44.04±2.42
Trait anxiety	42.41±1.45	42.90±1.72	41.00±2.74

Data are reported as mean more and less standard error of the mean

**Table 7:** Results of the follow-up

	All patients (N=96)	FARP (N=71)	Non-FARP (N=25)
Pain free	15	8	7
Pain improved	38	27	11
Pain unchanged	41	35	6
Pain worsened	2	1	1

**Table 6:** Depression and anxiety scores for male and female FARP and non-FARP patients

	All patients (N=96)		FARP (N=71)		Non-FARP (N=25)	
	Male (N=34)	Female (N=62)	Male (n=21)	Female (n=50)	Male (n=13)	Female (n=12)
Depression	10.59±0.69	14.65±0.95	11.10±0.93	15.58±1.12	10.08±0.78	11.58±1.19
State anxiety	43.94±2.56	45.27±1.90	45.19±3.59	45.02±2.21	41.92±3.44	46.33±3.43
Trait anxiety	40.79±2.67	43.29±1.71	43.57±3.48	42.62±1.98	36.31±3.97	46.08±3.30

Data are reported as mean more and less standard error of the mean.



Our study found that depression is common among patients with chronic anal pain. Long-term pain can inevitably cause psychological disturbance. Depression is more severe in patients with FARP as some researchers reported that depression occurs in about 30% of FARP patients [25, 26]. Since depression and anxiety were assessed at the time of pain diagnosis, we were unable to draw a conclusion whether pain precedes psychological disturbance. The causal effect between pain and psychological disturbance needs to be determined in the future studies. However, it was previously conveyed that FARP is affected by behavior and psychological factors [27].

In general, pain treatment for FARP patients is far challenging than the non-FARP patients because FARP has no obvious underlying anorectal or endopelvic organ disease symptom but it is a syndrome of idiopathic multi-factorial vague disorder resulting from a complex interaction among neurological, musculoskeletal and endocrine systems [27, 28]. In contrast, non-FARP patients usually have a clear cause for their pain and targeted treatment for the cause typically lead to good treatment outcomes.

Among common therapies for FARP such as electrogalvanic stimulation, biofeedback training, digital massage of the Levator Ani muscles, and Sitz bath, biofeedback training is considered as the most effective [29-32]. Still, a large percentage of patients do not respond to any pain treatment. In a study with 80 cases of FARP patients, Gary K [26] found that only 46.3% of patients had reduced pain after treatment, while the majority did not respond to the treatment. Perhaps combination therapies will reach higher treatment efficiencies. In a study of patients with the Levator Ani syndrome, most patients reported pain relief after a combination of massage, Sitz bath, muscle relaxants, and diathermy [33, 34].

In conclusion, chronic anal pain is a clinical manifestation stemming from diverse pathological and physiological changes. Although the examination in the urinary and reproductive system and other surrounding organs is needed to reach a definitive diagnosis, clear understanding of the underlying pathophysiology and the patient's psychological condition may help improve pain treatment.

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