1130-0108/2005/97/7/491-496
Revista Española de Enfermedades Digestivas

Rev Esp Enferm Dig (Madrid)
Vol. 97. N.° 7, pp. 491-496, 2005

Sequential treatment for proctalgia fugax. Mid-term follow-up

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ABSTRACT

Introduction: proctalgia fugax (PF) is a benign, self-limiting disease characterized by episodes of intense anorectal pain at frequent intervals in the absence of organic proctological disease. Even though PF was described more than a century ago, its etiology remains unclear. Currently there is no information available. Few papers quoting many ways of management have been published. The aim of this study was to investigate patients complaining of this condition and to treat them with sequential therapy.

Patients and methods: we devised a descriptive, prospective study of patients complaining of acute perianal pain –duration less than 30 minutes– without organic disease or previous perianal surgery since 1996 to 2002 in our Department. We treated these patients using a three-step treatment (1: information, hip bath, benzodiazepines; 2: sublingual nifedipine 10 mg, or topic 0.1% nitroglycerin on demand; 3: internal anal sphincterotomy if hypertrophy of the internal anal sphincter was demonstrated by anal ultrasonography and no improvement was confirmed with the previous steps of treatment). We defined remarkable improvement as a decrease in the number of episodes by half or in pain intensity by 50%.

Results: Fifteen patients with an average follow-up of 4 years. Anal endosonography confirmed a grossly thickened internal anal sphincter (IAS) in 5 cases. After the first step of treatment 7 patients improved and 1 patient was cured; after the second step of treatment 3 patients improved and 1 was cured; the third step was applied to 3 patients with a thickened IAS; 1 patient improved and 1 patient was cured.

Conclusion: a total resolution of PF is not always possible, but we may improve symptoms and their frequency. Almost 50% of patients in our series improved with the first step of treatment; 30% of our patients had IAS hypertrophy. Anal endosonography can help in the diagnosis of organic diseases or IAS hypertrophy, for which we can perform an internal anal sphyncter myectomy.

Recibido: 29-11-04.
Aceptado: 15-02-05.

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Key words: Anal pain. Proctalgia fugax. Sphincter hypertrophy. Sphincterotomy.

Gracia Solanas JA, Ramírez Rodríguez JM, Elía Guedea M, Aguilella Diago V, Martínez Díez M. Sequential treatment for proctalgia fugax. Mid-term follow-up. Rev Esp Enferm Dig 2005; 97: 491-496.

INTRODUCTION

Anorectal and perineal pain has been described in association with different pathologies that are usually easy to recognize, including hemorrhoids, fistula, fissures, perianal sepsis and carcinomas. But perianal pain can also be present in some cases where an organic cause cannot be found, and its pathophysiology remains uncertain. Proctalgia fugax had been traditionally included amongst other functional disorders that produce anorectal and perineal pain without recognizable organic disease, including coccygodynia, levator ani syndrome, vulvodynia, and perineal neuralgia. Although proctalgia fugax was described more than a century ago, its etiology is still unclear. Proctalgia fugax is a benign, self-limited disease characterized by episodes of deep recurrent anorectal pain. Even though nearly 14% of the population may suffer from proctalgia fugax, most patients do not seek medical advice (1). The diagnosis is based upon its particular clinical pattern and the lack of organic disorders explaining anorectal pain. Many therapies have been used in the management of such cases, often in an empirical way.

There are in fact few papers on proctalgia fugax in the literature, most of them focusing on the study of a particular treatment usually in a short series of patients.

The aim of this study was to prospectively study a group of patients complaining from this pathology, and to treat them according to a pre-established sequential therapy.

MATERIAL AND METHOD

From January 1996 to January 2002 all patients visiting our outpatient clinic because of proctalgia fugax were prospectively included in a specially designed data file. A detailed medical history as well as a perianal and rectal examination were performed for all cases. Anuscopy, rectoscopy and anal endosonography (AUS) were mandatory complementary diagnostic tests.

Criteria for inclusion in our study were: abrupt deep pain limited to the anal canal, with a maximum duration of 30 minutes and at least one episode per month.

Patients with synchronic organic lesions or with a history of surgery in the perianal region were excluded.

After an exhaustive MedLine research a therapeutic protocol was established based on three steps according to the most relevant bibliography (Table I). Following our protocol patients were offered the next step of treatment when no improvement was obtained with the previous.

Table I. Sequential treatment

1st step	Reassure and inform the patient Hip baths Oral benzodiazepines
2nd step	Sublingual nifedipine 10 mg or topic 0.1% nitroglycerin during crises
3rd step	Internal anal sphincterotomy

If no improvement was confirmed after 3 months of treatment, we moved to the next step if the patient agreed.

First step: reassurance about the disease, hip bath, benzodiazepines; second step: sublingual nifedipine 10 mg (or topic 0.1% nitroglycerin ointment if previous hypotension) at the time of episodes. Third step: internal anal sphincterotomy when AUS showed hypertrophy (IAS thicker than 3.5 mm) (2). The adequacy of the sphincterotomy was assessed by AUS at the first postoperative control.

All patients were asked to complete an analogical visual pain scale (VPS: 0=no pain, 10=worst pain) at the time of the crisis, and to point out the result on a diary.

We defined a remarkable improvement if the number of episodes decreased to a half or the level of pain (according to the VPS) dropped by more than 50%.

The follow-up was planned at 1, 3 and 6 months and yearly thereafter. Patients were contacted by phone in order to learn their current situation.

RESULTS

Fifteen (11 women) patients with a mean age of 46 years (range 36-60) could be included in our study. Average follow-up has been of 4 (range 2-6) years.

Symptoms are shown in table II.

Table II. Symptoms

Anorectal pain:	–Sudden onset –Variable character: sharp, gripping, cramp-like –No radiation
Self-limited:	–Duration: seconds to minutes
Recurrent:	–Often occurs at night –May occur 3 to 4 times weekly

The characteristics of pain (Table III) were: nocturnal pain in 73.3% (11 cases) of cases, with VSP reaching an average pain level of 8 (range 6-9). The duration of symptoms was 17 months (range 4-42) on average. The number of crisis was 2 per week (range 1/day -1/month).

AUS showed IAS hypertrophy in 5 patients.

Table III. Characteristics of pain

Average duration of symptoms:	17 months (4-42 m)
Anal pain severity (SVS):	8 (6-10)
Average pain duration:	15 min (5-20)
No. of crises:	2 week (1/day–1/month)
Onset of pain:	At night (73.3%)
IAS hypertrophy:	5 patients (33.3%)

After the first step in our sequential treatment, all patients improved (anal pain decreased to a level of 5.4; range 3-8) but just 7 of them fulfilled our criteria of remarkable improvement (anal pain average: 3.4; range 3-4; average number of crisis 1 per week), and 1 patient was cured. According to our protocol, the second step of treatment was offered to 7 patients (5 of them received sublingual nifedipine and the other 2 nitroglycerin ointment). Again, all patients improved (anal pain intensity level dropped to 4; range 2-7; average number of episodes 3 per month), but only three of them satisfying our criteria of remarkable improvement (anal pain average: 3). One patient was free of disease after this second step. As undesirable effects, 1 patient (20%) complained of mild headache and 2 patients (40%) presented facial flushing shortly after the nifedipine pill; such cases did not required to suspend the therapy. None patient under nitroglycerin ointment reported side-effect. In 3 cases with a demonstrated IAS hypertrophy and no improvement in previous steps, an anal internal sphincterotomy

was performed. After this third therapy step, one patient improved, with an intensity level of anal pain of 4. The other two cases, healed (Table IV).

Currently, 4 out of 15 patients are free of symptoms, 8 patients have some mild and sporadic crises (1 in 6 months), and only 3 patients remain complaining of recurrent acute anal pain (1/month).

Table IV. Improvement of symptoms/healing

	No. of patients	*Improvement*	*Healing*
1st step	15	7	1
2nd step	7	3	1
3rd step	3	1	2

DISCUSSION

Proctalgia fugax is a symptomatic, unstable disease with differences in the duration and recurrence of episodes, which makes it hard to include these patients within an homogeneous group. It is a very prevalent disease, but fortunately most patients have sporadic crises (1-2/year or even less) and do not demand medical advice (1). Patients seen in our outpatient clinic had a mean of 2 episodes per week. In fact, we could not use the number of crisis as an exclusion criterion.

Although the clinical appearance of proctalgia fugax was established almost a century ago, its etiology remains unclear. Moreover, due to a lack of proper studies, the management of these patients is controversial.

Some theories have appeared trying to explain the pathophysiology of proctalgia fugax: alterations in the internal anal sphincter function and its morphology (3). In fact, a hereditary form with sphincter hypertrophy has been identified of late (4,5). Some other cases have been reported in association with irritable bowel syndrome (6). Uncontrolled serotonine release would result in excessive hypermotility as well as in visceral hypersensitivity (7). Finally, other theories suggest that proctalgia fugax is caused by pshycological factors (8).

The diagnosis is based upon clinical manifestations and the absence of an organic origin for pain. Physical examination, rectoscopy, and anuscopy are normal in these patients. Endoanal ultrasonograhy or magnetic resonance imaging scans are usually normal, but sometimes may find an unexpected hypertrophy of the IAS; in such cases an internal anal sphincterotomy (as is the case in the hereditary form) may be of help. Anorectal manometry can demonstrate an increased IAS pressure with paroxysmal inability to relax in some cases (3).

The differential diagnosis of anorectal pain must include common organic diseases such as hemorrhoids, anal fissure, perineal fistula and sepsis, carcinoma, compression of sacral nerves (8), and gynecological disease (9).

Many treatments have been attempted with different success rates: cholinergic drugs (10), calcium antagonists (11), botulinum A toxin (12), adrenergic drugs (13) such as salbutamol inhalations during episodes, intravenous lidocaine (14), hip baths (15), topical nitroglycerin ointment (16), biofeedback techniques, anal dilatation, internal lateral sphyncterotomy when IAS hypertrophy is demonstrated (4,5), and psychotherapy (17).

At first glance ours may seem a short series, but in talking about proctalgia fugax it is by no means worthless. Moreover, all of our patients followed the same sequential treatment for at least 2 years. Having in mind the limited number of patients we only could devise a descriptive study, even if we knew that a better study would have been a case-control trial. For this reason we could not calculate statistical significances, and our results cannot be generalized. Healing is not always possible, but symptoms can be partly relieved and the rate of episodes reduced. Almost 50% of patients in our study were satisfied after the first step of treatment; in such cases no further treatment was necessary. We can conclude that one of the most important facts in the management of proctalgia fugax is to reassure patients about the disease, and many of them will learn to cope with their pain and to tolerate it better. In our series, 5 out of 15 (33.3%) patients had IAS hypertrophy on AUS. In 3 of them we carried out a sphincterotomy, which improved symptoms. Thus we advocate (with the limitations entailed by the number of patients) for anal endosonography as a useful tool in proctalgia fugax, first to rule out organic disease and secondly to identify patients with IAS hypertrophy who may benefit from sphincterotomy when conservative therapy fails.

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