Paroxysmal anal hyperkinesis: a characteristic feature of proctalgia fugax

S S C Rao, R A Hatfield

Abstract

***Background and Aims*—Proctalgia fugax is a common problem, yet its pathophysiology is poorly understood. The objective was to characterise colorectal disturbances in a paraplegic patient with a 10 year history of proctalgia fugax that began two years after an attack of transverse myelitis.**

***Methods*—Standard anorectal manometry and prolonged 33 hour ambulatory colonic manometry at six sites in the colon were performed together with myoelectrical recording of the anus. Provocative tests designed to simulate psychological and physical stress and two types of meals were included.**

***Results*—Anorectal manometry showed normal internal sphincter tone and normal rectoanal inhibitory reflex but an inability to squeeze or to bear down or to expel a simulated stool. Rectal sensation (up to 360 ml inflation) was absent. Pudendal nerve latency was prolonged (4·5 ms (normal <2·2 ms). During colonic manometry, the patient reported 27 episodes of pain, of which 23 (85%) were associated with bursts (1–60 min) of a high amplitude (0·5 to >3·2 mv), high frequency (5–50/min) anal myoelectrical activity, particularly after stress tests, meals, and at night. The myoelectrical disturbance only occurred with proctalgia. Intermittently, 16 bursts of 3 cycles/min phasic rectal contractions were seen, but only six were associated with proctalgia. Colonic motility was reduced compared with normal subjects.**

***Conclusions*—The temporal association between a high amplitude, high frequency myoelectrical activity of the anal sphincter, and the occurrence of proctalgia suggests that paroxysmal hyperkinesis of the anus may cause proctalgia fugax.**

(*Gut* 1996; **39:** 609–612)

Keywords: proctalgia fugax, manometry, colon motility, pathophysiology.

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Accepted for publication
9 May 1996

Proctalgia fugax affects 14% of healthy adults.[1 2] It is described as a sudden, severe, and episodic anorectal pain that often radiates to the gluteal region and occurs at night.[1–3] Its pathophysiology remains unclear.[1–10] It has been hypothesised that proctalgia fugax may arise either from spasm of the rectal[4 5] or the pubococcygeal muscle,[6] but a specific anomaly has not been found. Harvey[7] suggested that phasic rectosigmoid contractions rather than levator ani spasm may cause proctalgia, although, in their study 42% of contractions were painless and anal motility was not recorded. Others have implicated stress.[8] More recently a hereditary myopathy of the anal sphincter has been described.[9 10]

We investigated the pathophysiology of proctalgia fugax by examining the anorectal and colonic motor function in a patient with paraplegia and proctalgia. This unusual problem provided a unique opportunity for assessing this condition. Our objectives were threefold: (1) Is it possible to reproduce proctalgia fugax? (2) Is proctalgia associated with a motor abnormality? (3) Where is the anatomical locus – anal or colorectal?

Case report

A 56 year old professor presented with a 10 year history of episodic anorectal pain. The pain was squeezing in nature, radiated to the gluteal region, and was severe enought to wake him every other day. Usually, it lasted 5–10 minutes and sometimes an hour. Precipitating factors included meals, stress, prolonged sitting, and digital anal stimulation.

Twelve years ago, after an attack of viral transverse myelitis that affected T9, he developed paraplegia. Simultaneously, he developed urinary retention with overflow and detrusor instability which required self catheterisation and ditropan (10 mg twice daily). He also developed constipation with inability to initiate defecation. This required laxatives and digital stimulation. Other medications included valium, baclofen infusion (50 mg/day) for muscle spasm, and amitriptyline (100 mg/day) for depression. Opiates, transcutaneous electrical nerve stimulation unit, clonidine, and digital massage were ineffective in treating proctalgia.

EVALUATION BEFORE STUDY

Anocutaneous reflex and perianal sensation were absent. The external anal sphincter was patulous but internal sphincter tone was normal. Haematology, electrolytes, serum calcium, and magnesium were normal. Anoscopy was normal. Colonoscopy disclosed a hypotonic colon with normal mucosa.

PROVOCATIVE TESTS

Tests of anorectal function

A manometric probe with five pressure sensors and a balloon was placed in the rectum. Pressure activity was recorded on a data logger

(Gaeltec-7 MPR, Isle of Skye, UK). Firstly, a series of manoeuvres were performed that included squeezing and simulated defecation.[9] Next, using intermittent balloon distension (10 to 360 ml), rectal sensation and rectoanal reflexes were evoked. Pudendal nerve latency was measured as described previously.[11]

Ambulatory colonic manometry

Forty eight hours after discontinuing baclofen and amitryptilline, the patient was admitted and received a tap water enema. Next, a solid state probe with four pressure sensors and one integrated EMG sensor was placed in the colon with the help of a colonoscope. The pressure sensors were located at 7, 14, 25, and 45 cm and the EMG sensor at 2 cm from the anal margin and their position was confirmed by fluoroscopy. No sedation was used. Pressure and myoelectrical activity were recorded for 33 hours with an 8 Hz portable recorder (Gealtec-7 MPR) with an EMG bandwidth of 15 Hz–1·5 kHz. Symptoms were recorded using an event marker and described in a diary. The study included the following tests.

Dichotomous listening test – Psychological stress was induced by a dichotomous listening task (one hour), preceded by a control phase (one hour) of listening to a narrative passage[12]; recovery (one hour) entailed listening to music. Pulse rate and blood pressure were recorded every five minutes.

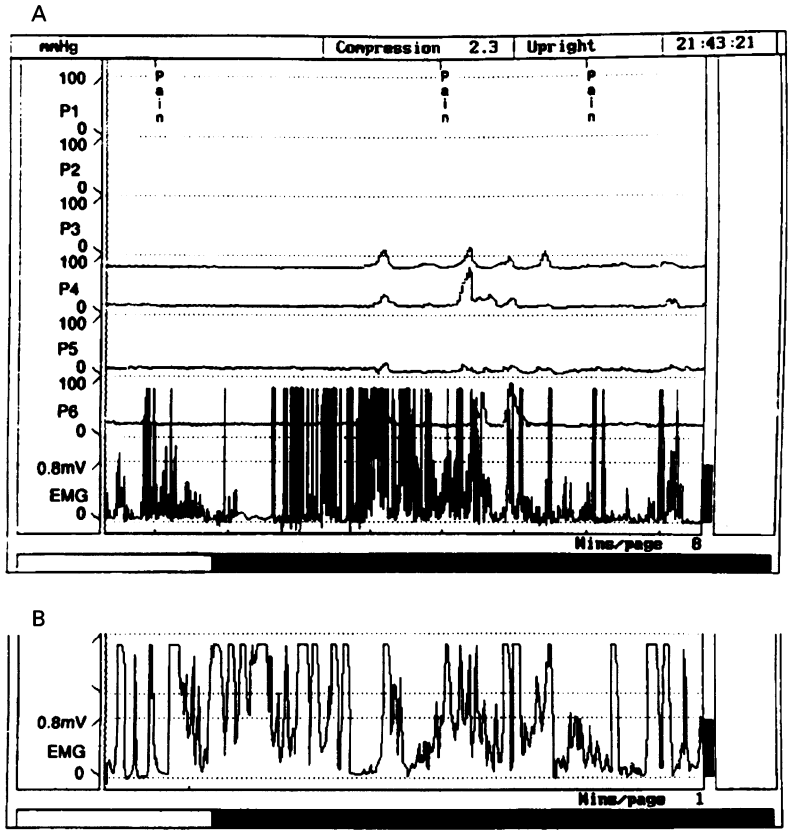
Cold water immersion test – This three phase test simulated physical stress, and consisted of immersing each hand in water at 37°C (control) and at 4°C (test) for 15 minutes. Recovery (30 minutes) entailed simple rest. Each hand was immersed for one minute and then held outside for 15 seconds. Pulse rate and blood pressure were recorded every five minutes.

Gastrocolonic responses – The patient was served two 1000 kcal meals; a high fat meal and a high carbohydrate meal.

TABLE I *Results of anorectal manometry and electrophysiology*

	Patient	*Normal subjects*
Maximum resting anal pressure (mm Hg)	75	67 (20)
Maximum squeeze pressure (mm Hg)	80	158 (38)
Threshold for first sensation (ml)	Absent	20 (8)
Threshold for desire to defecate (ml)	Absent	73 (28)
Pudendal nerve terminal latency (ms)	4·5	2·0 (1·1)
Rectal compliance (dv/dp) at 100 ml (ml/mm Hg)	20·0	12·5 (3·2)

Results for normal subjects are mean (SD).



(A) Typical example of colorectal motility tracing during an episode of proctalgia fugax. The sensors were located as follows: P_3 at 45 cm, P_4 at 25 cm, P_5 at 14 cm, P_6 at 7 cm, and the integrated EMG sensor at 2 cm. The EMG channel displays high amplitude, high frequency myoelectrical spike activity that was associated with proctalgia fugax. The pressure sensors show isolated random contractions in the distal colon. (B) Example of the anal hyperkinesis at a slower speed (1 min/page).

Data analysis

Anorectal manometry – Resting and squeeze sphincter pressures, rectal and anal pressures during simulated defecation, rectoanal reflexes, rectal compliance, thresholds for rectal sensation[9] and pudendal nerve latency were measured.[11]

Ambulatory colonic motility – The manometry was analysed to identify any association between symptoms and manometric events. Additionally, during the stress tests and during the one hour preprandial and postprandial periods, pressure waves with an amplitude >8 mm Hg and a duration >3 seconds were identified and their mean frequency and area under pressure waves were compared with those of healthy subjects.[13]

Results

ANORECTAL TESTS

Resting sphincter pressure was normal, external sphincter was weak and the pressure did not change with squeeze or straining (Table I). Rectoanal inhibitory reflex was present. Rectal sensation was absent. Rectal compliance was higher. Pudendal nerve latency was prolonged (Table I).

AMBULATORY COLORECTAL MOTILITY

During this test, the patient reported 27 episodes of typical pain, of which 23 (85%) were associated with unusual myoelectrical activity of the anus (Figure) with an amplitude of 0·5 to >3·2 mV and a frequency of five to 50/minute. The duration of each episode varied from three to 60 minutes. On three occasions the patient woke up with pain and two of these were associated with anal hyperkinesis. The anal myoelectrical disturbance did not occur in the absence of proctalgia. Intermittently, 16 bursts of phasic rectal contractions at 3 cycles/minute were seen.[16–18] Six of these occurred during an episode of pain.

TABLE II *Effects of physiological and physical stress on colonic motor activity*

	Control		*Stress*		*Recovery*	
	Patient	*Normal subjects*	*Patient*	*Normal subjects*	*Patient*	*Normal subjects*
Psychological stress:						
Pulse rate/min	80	68 (11)	84	78 (9)	68	68 (8)
Blood pressure (mm Hg)	143/63	110 (12)/68 (9)	157/74	111 (10)/70 (7)	141/62	113 (9)/69 (9)
No of waves	67	51 (39)	70	74 (43)	43	69 (31)
AUC×10^3	5·3	4·8 (2·4)	7·3	7·7 (4·2)	4·9	8·1 (3·9)
Physical stress:						
Pulse rate/min	65	73 (10)	79	74 (11)	62	67 (11)
Blood pressure (mm Hg)	137/57	114 (11)/69 (10)	173/78	130 (20)/79 (14)	158/64	118 (15)/71 (9)
No of waves	23	52 (18)	28	71 (34)	34	52 (28)
AUC×10^3	1·4	5·4 (2·4)	3·3	6·9 (3·4)	3·2	6·0 (3·0)

Values are mean or mean (SD). AUC=area under curve.

EFFECTS OF PSYCHOLOGICAL AND PHYSICAL STRESS

During the stress phase, the patient reported frequent episodes of pain that were associated with anal hyperkinesis. When compared with normal subjects there was decreased motility, but the autonomic responses were similar (Table II).

EFFECT OF MEALS

Both meals increased colonic motor activity and induced proctalgia, but compared with normal subjects the patient's colon showed less activity (Table III).

Discussion

We investigated the possibility that proctalgia fugax was associated with a motor dysfunction of the anorectum. Anorectal manometry showed changes that were consistent with those reported previously after high spinal anaesthesia[14] and high spinal cord injury.[15] However, our patient was unique in that he reported proctalgia, although he had pudendal neuropathy and objective testing disclosed absent somatic and visceral anorectal sensation. Because proctalgia fugax is a paroxysmal condition, it was not surprising that a motor abnormality was not seen during the short manometric study. Hence, a prolonged colorectal manometry was performed.

During this study, the patient reported discrete episodes of proctalgia that were temporally associated with a unique motor disturbance, characterised by spontaneous, high amplitude, high frequency myoelectrical activity of the anus. This has not been shown previously. This pattern was invariably seen during painful episodes and at night when the patient woke up with pain, but was never seen in the absence of pain. Recently, endosonography showed fluctuations in diameter of the anal sphincter in a patient with proctalgia.[10] Our study not only confirms but extends this finding by providing an objective demonstration of the anal myoelectrical disturbance. Furthermore, it provides corroborative evidence for paroxysmal anal hyperactivity as a mechanism for proctalgia.

It has been suggested that rectosigmoid contractions may cause proctalgia.[5 7] Although we noted 16 bursts of phasic rectal contractions during the study, two thirds of these were not associated with proctalgia. Moreover, a 3 cycles/minute phasic activity is a characteristic feature of the normal, healthy rectosigmoid colon.[16 17] Hence, it is unlikely that phasic rectal contractions cause proctalgia fugax. Our study suggests that the anal sphincter is the possible locus for this problem.

The internal anal sphincter is regulated by tonic excitatory discharge from the thoracolumbar sympathetic nerves.[14 18] Because the sympathetic innervation was probably intact in this patient (supported by normal autonomic responses), it is conceivable that the paraoxysmal hyperkinesis of the anal sphincter was possibly mediated through this system. By contrast, the external anal sphincter is innervated by the pelvic and pudendal nerves and is regulated by the central nervous system. The myelitis may have disrupted this pathway causing the patulous external anal sphincter. The myelitis may also have exacerbated the proctalgia by facilitating uninhibited sympathetic discharge to the anus.

The incidence of proctalgia was higher after the provocative tests confirming the patient's history. However, the increase in motor activity after stress tests and after meals was less than that of normal subjects, although the autonomic responses were comparable. The attenuated motor responses were probably due to the hypotonic colon.[19]

This study shows that prolonged ambulatory recording of anorectal motility may serve as a useful diagnostic test for patients with proctalgia fugax. Whether our findings are peculiar to a paraplegic patient with this problem or are a common disturbance that has not been previously recognised merits further investigation. Our study shows that proctalgia fugax is associated with a paroxysmal, high amplitude, myoelectrical disturbance of the anal sphincter, which may be due to an

TABLE III *Effects of a fat and a carbohydrate meal on colonic motor activity*

	Preprandial		*Postprandial*	
	Patient	*Normal subjects*	*Patient*	*Normal subjects*
Fat meal:				
No of waves	32	109 (71)	89	141 (58)
AUC×10^3	4·7	12·6 (13·4)	12·3	18·5 (10·5)
Carbohydrate meal:				
No of waves	57	108 (46)	100	161 (61)
AUC×10^3	5·6	9·8 (4·6)	10·5	17·8 (9·4)

Values are mean or mean (SD).

overactivity of the sympathetic discharge to the anal sphincter and could explain why symptoms occur durings stress.[2 8]

This research was supported in part by the AGA SKF-Beecham Clinical Research Award, and by Grant RR00059 from the General Clinical Research Centers Program, National Center for Research Resources, National Institutes of Health.

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