Ergotamine-induced solitary rectal ulcer

Ofer Shpilberg¹, Michael Ehrenfeld¹, Dov Abramowich¹, Yechezkel Samra¹ and Leon Bat²

¹The Rusinow Department of Internal Medicine C and ²the Institute of Gastroenterology, The Chaim Sheba Medical Center, Tel Hashomer and The Sackler School of Medicine, Tel Aviv University, Israel.

Summary: A rare case of ergotamine-induced solitary rectal ulcer is described in a 41 year old woman who used high doses of ergotamine tartrate-containing suppositories for severe migraine headaches. Complete recovery of the ulcer was noticed after cessation of treatment with the suppositories. The relevant literature is discussed.

Introduction

Solitary ulcer of the rectum is a rare entity with a wide spectrum of clinical manifestations which has lead to it being called the solitary rectal ulcer syndrome (SRUS).¹ This term can still be potentially misleading, since the endoscopic appearance may be neither solitary nor ulcerated.² A considerable uncertainty still exists as to the cause, natural history and management of this uncommon condition.

In recent years, a few cases have been reported describing a new entity termed by some as ergotamine colitis^{3,4} or by others anorectal ergotism.⁵ This rare condition bears some features similar to that of the SRUS. However, some characters of this entity are clearly different from the SRUS to such an extent that make it a separate entity.^{3–5} We describe a case of ergotamine-induced solitary rectal ulcer and review the previously reported cases.

Case report

A 41 year old European female presented herself with a 2-year history of abdominal pain, irregular bowel movements and rectal discomfort. Since the age of 19 she had suffered from migraine-type headaches, and during the last 2 years, she noticed a marked aggravation in the frequency and intensity of these headaches. She had therefore started to use various kinds of analgesic rectal suppositories, including a weekly consumption of 14–21 supposi-

tories of ergotamine tartrate, each of them containing 2 mg of the substance and 100 mg of caffeine. Concomitantly she started to suffer from non-colicky, continuous abdominal pain, irregular bowel movements and rectal discomfort, with extreme difficulty in defecation. No other gastrointestinal signs or symptoms were noticed.

Upon examination, she was found to be in good general health, and the only abnormality was a bulky irregularity of the rectal mucosa some 6–7 cm from the anal verge. Gynaecological and neurological examinations were normal.

Laboratory tests including complete blood count, liver and kidney function tests were all normal. Stool examinations were negative for amoeba, shigella and salmonella. Colonoscopic examination revealed a solitary ulcer with sharp edges, 6–7 cm from the anal orifice in the left anterior aspect. No other pathological findings up to the level of the caecum were found. Histological examination of the ulcer edges revealed acute and chronic proctitis with inflammatory exudate and granulation tissue, inflammation of lamina propria with polymorphonuclear cell infiltration irregularity of the glands of Luberkin with large spaces in between them, oedema and haemorrhages. Further investigations, including abdominal and brain computed tomographic scans and liver scintigraphy, did not show any abnormal findings.

The patient was diagnosed as suffering from solitary rectal ulcer which was attributed to ergotamine tartrate-containing suppositories.

Four weeks later after cessation of the treatment with these suppositories a follow-up rectoscopy showed complete recovery with disappearance of the rectal ulcer, and a concomitant improvement in the patient's complaints. Eight months later a follow up colonoscopy was completely normal.

Discussion

The case described herein (case number 9 – Table I) is, to the best of our knowledge, the ninth reported in the English language, of a new entity termed solitary rectal ulcer due to anorectal ergotism.^{3–5} All patients used rectal suppositories containing ergot preparations for migraine-type headaches. Seven out of 8 (87.5%) were women (Case number 1 in Table I was excluded because of lack of data) with a mean age of 47.5 years (range 41–57 years). These patients used the ergot-containing suppositories in an average dose of 27.8 mg/week (range 4.5–63 mg/week) which is above the recommended dosage for use (10 mg/week). All patients in this series used the suppositories for at least one year with an average of 3.25 years (range 1–8 years). Usual symptoms included anorectal pain, rectal bleeding, tenesmus, diarrhoea and irregular bowel movements. In 5 patients the lesions were confined to the rectum, in 2 to the anus, and in the remaining 2 there were combined lesions both in the rectum and the anus (including Case no. 1). The mean recovery time after cessation of therapy was about 7 weeks (range 2–10 weeks). Single reports of this entity have also been published in other languages.^{6–8}

The pathogenesis of the above mentioned entity may be explained in two ways. First, high doses of ergot-containing suppositories may cause local vasoconstriction which results in mucosal ischaemia and ulceration. It is known that ergotamine

toxicity can manifest in a variety of adverse effects due to systemic vasoconstriction, including acute ischaemia of extremities, acute myocardial infarction and sudden death.^{9–11}

It still remains unclear why the possible local vasoconstriction is confined to the site of the suppository absorption, and does not produce systemic vasoconstriction in other sites. It is known, however, that ergot alkaloids exert not only a general constrictive effect on the arterial tree, but also a venoconstrictor effect.¹² Since the suppository is being absorbed slower than expected, a local effect on the epithelium and the vascular tree may be seen.

An alternative possible mechanism of ergotamine-induced rectal ulcer is that ergot-containing suppositories abuse can lead to direct mechanical pressure resulting in decubitus ulcer. A combination of the above two mechanisms is also possible.

It is still unclear whether the ergotamine-induced ulcer is part of the SRUS or a distinct entity by itself. Endoscopic and histological findings cannot differentiate between the two disorders. The lesions are sharply circumscribed and biopsy specimens reveal infiltration of the lamina propria by polymorphonuclear leukocytes and fibrosis, surrounding areas of necrosis.⁵

The two entities are different in their aetiology and clinical course. There is no known aetiology to the SRUS and the symptoms and endoscopic findings may persist for years no matter what treatment is used,^{5,13} while all the patients with the

Table I Clinical data of patients with ergotamine-induced solitary rectal ulcer

Case no.	Age	Sex	Symptoms	Endoscopic findings	Maximal dose of ergotamine mg/week	Duration of ergotamine abuse (years)	Recovery period after ergotamine discontinuation	Reference
1	Unknown	Unknown	Unknown	Rectal ulcer	Unknown	Unknown	Few weeks	3
2	45	F	Diarrhoea tenesmus	Rectal ulcer	63	6	2 weeks	4
3	44	F	Anorectal pain	Rectal ulcer	16	2	Few weeks	5
4	49	F	Rectal bleeding	Rectal and anal ulcer	10	8	4 months	5
5	50	F	Rectal bleeding	Rectal ulcer	12	2	2 months	5
6	57	F	Anorectal pain	Rectal and anal ulcer	4.5	1	6 weeks	5
7	44	F	Rectal bleeding	Anal ulcer	60	4	4 weeks	5
8	50	M	Rectal pain	Anal ulcer	15	1	4 weeks	5
9	41	F	Rectal pain Abdominal pain Irregular bowel movements	Rectal ulcer	42	2	4 weeks	Current report

ergotamine-induced colitis have a strong history of ergotamine-containing suppositories abuse, and all the symptoms and endoscopic findings disappear usually within 7 weeks of discontinuation of the

suppositories (Table I). Future observations may contribute to the understanding of this unique entity.

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