Effective treatment of solitary rectal ulcer syndrome using argon plasma coagulation

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Abstract

Solitary rectal ulcer syndrome (SRUS) is a chronic, multiform, non-cancerous disorder of the rectum, the final diagnosis of which is based upon histopathological criteria. This disorder is often accompanied by latent proctoptosis. We present a patient who (in 1996) was the first case in which argon plasma coagulation (APC) was used for SRUS treatment. In the years 2004–2005 the same patient underwent 15 APC sessions (at monthly intervals) obtaining full recovery from SRUS, although she had been treated unsuccessfully for 17 years prior to that. Six-year observation did not show any relapse. Local therapy with APC seems to be an important alternative in SRUS treatment without prolapse of the rectum and could become a basic method for bleeding treatment in SRUS.

Introduction

Solitary Rectal Ulcer Syndrome (SRUS) is a chronic, non-cancerous disease of the rectum, occurring in the form of ulceration, a polyp, or a flat lesion. In SRUS diagnosis one should consider the clinical symptoms and the course of the disease, supplemented by the results of defecography, anorectal manometry, and endoscopic biopsies. The final diagnosis is set on the basis of histopathological criteria. It is often accompanied by evident or latent proctoprosia, and in cases like this the therapeutic possibilities and prognosis are slightly better.

The SURS is a rare disease, with an estimated incidence of 1–3.6 cases/100,000 people. In most observations, differences between genders are minor but it is reported in a slightly higher proportion of women. Although cases in children and elderly people have been reported, the disease mostly occurs in the third decade of men's life and in the fourth decade among women [1–4].

The diagnosis of SRUS is usually late, mainly because of its common asymptomatic initial course [1, 2, 5]. Later symptoms are the same as in other – more common – disorders of the anorectum and are often ignored. Treating SRUS is difficult and includes a variety of strategies, starting with conservative methods up to various surgery techniques. Despite numerous articles on SRUS and relatively large interest in the disease, the pathogenesis is still not known precisely and no effective treatment of solitary rectal ulcer syndrome has been established.

The aim of the study is to present an effective treatment of solitary rectal ulcer syndrome without proctoptosis with the pioneering use of argon plasma coagulation (APC).

Case report

A female, 62-year-old patient, (presently 79 years old) was admitted in 1994 to the University Ward of General Surgery and Gastroenterology in Bytom, Silesian Medical University for diagnosis and treatment of rectal ulceration.

The interview and documents showed that since 1988 the patient had disorders in the form of idiopathic, acute pains of the stomach, in the region of the anus and rectum, and diarrhoea alternating with constipation, mucus and blood in stools and decreased body mass. The colonoscopy (performed in another hospital) 10 cm from the anus showed the presence of rectum ulceration. Histopathology of a specimen was as follows: *Adenocarcinoma bene differentiatum exulceratum*. As a result, in 1989 the patient had exploratory laparotomy performed, which did not show any pathologic changes in the abdominal cavity. Subsequently, the patient used conservative treatment in the form of a diet. She felt quite well and did not report serious pain or constipation.

In 1992, however, the pain appeared again. The patient was still treated by conservatory treatment plus sulphasalazine and neomycin enemas, and control colonoscopies with samples taken for histopathological examinations were also carried out. However, each time the presence of rectal ulceration covered with fibroma with tiny polyp lesions on its edges was shown, the primary diagnosis of neoplastic change was never confirmed again. In the years 1992–1994, seven colonoscopies were done, which did not show any significant macroscopic and size changes of the ulceration.

As well as exploratory laparotomy done in 1989, she had an appendectomy in 1957 and a hysterectomy with perineoplasty in 1983. At that time the diagnosis was as follows: *Uterus myomatosus. Ruptura perinei inveterate*. The histopathology results were as follows: *cervicitis chronica, hyperplasia polyposa endometrii, cystis lutеica haemorrhagica, salpingitis*. In 1993 she had a benign ischaemic cerebral stroke.

The patient's mother had died due to cancer of the large intestine.

At admission the patient did not report any pain; the reason for admission was the presence of large amount of blood and mucus in stools. The patient was in good general condition. Apart from obesity (body mass index (BMI) 29.59 kg/m^2, height 156 cm, weight 72 kg), there were no deviations from normal condition. Rectoscopy and colonoscopy with samples for histopathology, panendoscopy, contrast infusion, hydrosonography, cystoscopy, and abdominal cavity USG were performed. The rectoscopy showed a 10-cm bleeding ulceration, and colonoscopy showed dolichocolon and two benign polyps, which were removed (in the descending colon – adenoma tubulare 41705 of 0.5 cm diameter and in the rectum on the 10 cm ulceration edge – polypus mucosus glandularis 41706). Contrast infusion showed numerous additional colon flexures, slight atrophy of large intestinal haustration and elongation of the transverse colon. Hydrosonography confirmed the atrophy of large intestinal haustration. Abdominal cavity ultrasound showed discreet urinary retention in the kidneys and cholecystolithiasis. No other deviations were observed. Laboratory tests done during hospitalisation showed anaemia – Hgb 11.9 mg%, Htc 35%, AST activity and glucose level in blood higher than the norm. The blood glucose curve was pathologic. The remaining tests showed no deviations. The local ulcer treatment by argon plasma coagulation with demonstrative appliance by ERBE – Erbotom ICC 200 with an APC attachment was suggested to the patient. Six APC sessions were done during colonoscopy every 2 days, with the result of ulceration decrease and no bleeding after the fifth session. The patient was dismissed in a good condition, with the recommendation of using high-fibre diet and outpatient check-ups. The patient did not follow the recommendations because at the time she had no pain or bleeding.

In March 1996 the patient was admitted to hospital due to recurrence of pain and considerable bleeding from the anus. Colonoscopy showed the presence of large ulceration surrounded by congested mucosa, covered with clots up to the sigmoid-rectal bend. The surgical procedure of transversostomy was performed, excluding the changed part of the rectum from the passage, and in the postoperative period, local treatment of ulceration with hydrocortisone enemas was applied. The subsequent retroscopies and colonoscopies showed slow regression of the lesions.

The colonoscopy done on July 15, 1996 showed that the ulceration had decreased by 70% compared to March 1996. The examination of May 1997 confirmed the lack of ulceration, so the decision to remove the transversostomy was taken.

In March 1998, which is 10 months after the reconstruction of the alimentary duct continuity, the patient was readmitted to the hospital due to rectal bleeding. The colonoscopy showed an irregular ulceration with bleeding, congested edges in the rectum on the front wall. The histopathology result (50891) revealed *ulceratio chronica*.

For the next 5 years control colonoscopies with ulceration samples for histopathology tests were done every year, and they confirmed the earlier diagnosis of *ulceratio recti chronica* – ulceration with tiny polyp lesions on the perimeter, with an average size of 2 cm × 3 cm. At that time, the patient did not suffer any pain, but periodically she had slight bleeding from the rectum. The patient was on a high-fibre diet.

In 2003, without any symptoms increase, the ulceration enlarged to the size of 3 cm × 4 cm. Histopathology (63830) showed *ulceratio chornica*. In July 2003, photodynamic colonoscopy was done in the Centre of Diagnostics and Laser Therapy. The results were as fol-

lows: in white light – a sitting polyp of 4–5 mm diameter of unchanged mucosa in the ascending colon, visible 7-cm, flat, irregular ulceration covered with fibrin with un-circumvallate edges in the rectum. In fluorescence light – ulceration edges locally of diffuse reddish fluorescence (congestion), 1 cm distally from the ulceration irregular brownish fluorescence field. Histopathology results (330858-861) were as follows: 1. ulceration edge – superficial mucosa fragments with the features of *inflammatio chronica non activa*. Epithelium sine dysplasia; 2. ulceration samples – superficial mucosa fragments. Epithelium without dysplasia; 3. ascending colon polyp – *tela adenomatis tubularis* I°. Dysplasia *epithelii glandularis* LG. Prolific inflammatory infiltrations, mainly eosinophilia.

In January 2003 the patient was offered argon therapy in our hospital. The patient suffered from diabetes mellitus, arterial hypertension, ischaemic heart disease, general atherosclerosis, degeneration of L-S spine joints, left hip and knee joints, cholecystolithiasis, and lower limbs varices. The patient had suffered ischaemic brain stroke with a slight left-side paresis and no distinct defect symptoms of CNS. Ankle/arm index (A/A) 0.7.

The patient was offered a method with the use of argon beamer by EMED, which had given promising treatment results so far; the recommendations of Stoppino *et al*. [6] were also taken into consideration. The method of local application was modified by extending the periods between argon coagulation, and the target was full healing of the ulceration. Fifteen APC sessions were done at monthly intervals, stopping the ulceration bleeding after the second session and decreasing the extensive ulceration constantly, and consequently obtaining full recovery from SRUS after 20 months of treatment. Further check-ups in a surgery clinic were recommended.

The patient is still under surgical supervision. The observation has currently been ongoing for 6 years, with no relapse. The latest colonoscopy was performed on Aug 10, 2010, during which the colonoscope was inserted into the caecum. The observations were as follows: the anus area with intertrigo; the intestine with numerous acute colon flexures, locally with flabby wall; in the rectum, 10 cm from the anus on the front wall, a maculate site was visible with 2.5 cm diameter and mucous membrane paler than the environment (at the site of previous ulceration) on a hard foundation – samples were taken for histopathology (the result of which showed superficial mucosa fragments of the large intestine).

Discussion

Patient noticed the first symptoms at the age of 56 years, which is later than the most common cases of SRUS in women (usually between 40 and 50 years; however, there is a wide age-range for the disease: 12–77 years) [1–4]. The SRUS aetiology, despite many studies and observations, has not been discovered. The most popular hypothesis claims that this is a secondary disease following rectal prolapse (overt or latent, mucosal or complete prolapse) and it is a result of defecation disorders. Disorders of muscle synergy related to defecation are seen in 25–82% of patients with SRUS [2]. Sharara *et al*. [1] estimate that the proportion of defecation disorders in patients is as high as 75%. Another hypothesis related to defecation disorders draws attention to the oversensitivity of rectal mucosa, leading to a continuous feeling of incomplete defecation and excessive rectal tenestmus [2]. The influence of atherosclerotic changes on mucosal ischaemia is also suggested [5], as well as disorders in cholinergic synapses of the autonomous nervous system, which can be related to diabetic polyneuropathy [7]. It is also suspected that SRUS can be an innate malformation of the hamartoma type.

There are other factors that are taken into consideration in the pathogenesis, such as an injury caused by a patient while initiating defecation with a finger or while using rectal enemas, the influence of radiotherapy, and the effects of some drugs applied per rectum. The coexistence of several factors in SRUS pathogenesis is also possible. A distinctive element that might be important in SRUS pathogenesis in this patient was the atherosclerotic component, dementia, diabetes mellitus and temporal relation with the perineoplasty done 5 years before. In this case, SRUS did not coexist with rectal prolapse, which considerably limits treatment options when conservative treatment is ineffective. The patient had diabetes mellitus, ischaemic disease, and hypertension. At the age of 62 she had already had a cerebral stroke with apparent dementia, which made introduction of a proper diet and effective psychotherapy difficult.

Lack of uniform agreements about procedures in SRUS also results from non-homogenous and poorly recognised pathogenesis. The therapy initially consists of conservative methods and is usually the end of therapy if there is some improvement or if patients are asymptomatic. Subsequently, it is recommended that patients follow a high-fibre diet to regulate defecation rhythm, perform relaxation exercises of the sphincter muscles, and avoid mechanical procedures leading to local injuries of rectal mucosa. Good results were achieved by using behavioural therapy: training based on biological feedback. This method is based on establishing control of the external rectal sphincter with relation to the patient's biological needs. The aim is to improve defecation technique, to decrease rectal over-

sensitivity to stimuli, to recover normal mechanism of defecation and thus to make clinical symptoms disappear, to normalise the frequency and quality of defecation, and to heal the lesions in the rectum. Most probably the feedback influences autonomic innervation and, as a result, improves mucous flow and provides favourable conditions for rectal wound healing [1, 7, 8]. The patient's education and behaviour modification are effective in cases of moderate symptoms and no substantial rectal prolapse. However, some authors claim that feedback is effective in short-term observations, but in the long-term perspective this method is ineffective. In our patient the only form of psychotherapy was the recommendation to follow the diet and to defecate regularly. It seems that the diagnostic histopathology mistake and probably the overly eager decision to perform a laparotomy did not influence the natural course of the disease.

It is usually a long time from the first symptoms to a diagnosis (from 3 months to 30 years, with a mean of 5 years) [1]. In our case, the diagnosis was established 6 years after the first symptoms. Late diagnosis is usually the result of ignoring symptoms typical for common anorectal diseases. The disorders usually include the presence of mucus, fresh blood, and/or mucous-purulent matter. Additionally, defecation disorders are common, including problems with initiation of defecation, painful, long-lasting rectal tenestmus, a feeling of incomplete defecation, and constipation [1, 4, 7, 8]. It is often connected with frequent visits to the toilet and initiating defecation using a finger. The SRUS is usually initially asymptomatic. In 26% of patients with SRUS, the only proof of the disease is from endoscopies and radiological examinations. Some patients with SRUS have mental, emotional disorders or are mentally retarded [1, 2, 7].

The final SRUS diagnosis is established on the basis of histopathological criteria. A study of samples taken at endoscopy shows changes in all the layers of the rectal wall: muscular, submucosal and mucous. The amount of collagen fibres is increased in all those layers. Each of them becomes thicker, especially the muscular membrane of mucous membranes. Mucous glands, which produce the excess of abnormal mucus, can be distributed incorrectly in submucous membrane. Moreover, there are anomalies of capillary vessels of mucous membrane, superficial erosions, and exudates [1, 3, 5, 8].

The lesions occur about 5–10 cm away from the anus fissure. They are of polymorphic character: ulcerations, polyp-like changes, and flat changes [3]. Polyp changes dominate in a non-symptomatic group, and ulcerations always seem to be symptomatic. Isolated ulcerations account for just 20% of all cases [1]. A typical ulcer is shallow, regular, and well separated. At the bottom of the ulcer there is white necrotic tissue, and its edge makes a ring of reddening and oedema. The diameter of the ulcer ranges from 0.5 cm to 4 cm, usually it is 1–1.5 cm. Macroscopic changes in SRUS are localised mainly on the rectal front wall, and more rarely on the dorsal and lateral wall. They can make up a large circuitous area [1, 4, 5]. Patient, while in the symptomatic period, had individual rectal ulceration flushing blood covered with clots on the front wall. In the non-symptomatic period, as well as ulceration covered with fibrin, small polyp changes were observed.

In 1994 a new unconventional therapy using APC was applied, the purpose of which was mostly to stop ulceration bleeding. The APC is a system of non-contact, unipolar coagulation, in which a stream of ionised gas is the energy carrier. Initially, it was used in classical surgery, and then, when the use of flexible endoscopes became commonplace, it was also used in interventional endoscopy for the treatment of malignant and benign tumours, and vascular malformations of the alimentary duct and breathing airways [9].

To treat SRUS we used a rented demonstrative appliance. As it turned out, the therapy had positive effects: it stopped the bleeding and improved the local condition. The APC therapy was continued up to the moment of bleeding cessation, but not to full recovery. It was decided to apply a conservative treatment in the form of a diet and check-up examinations. The relapse of bleeding and pain, and significant enlargement of the ulceration in 1996 made us perform a temporal colostomy, which is a final and very radical treatment option. Additionally, hydrocortisone enemas were applied in the postoperative period. When SRUS is accompanied by rectal prolapse, the aim of the treatment is surgical removal or fixation, sometimes parallel to lesion elimination. However, the biggest problem is the treatment of SRUS patients without rectal prolapse [2, 4].

The only solution, apart from the exclusion of the rectum from the passage, which is not very effective and burdened with complications, is surgical removal of the lesion. This is especially difficult when the ulceration is large [2, 4]. However, there are some reports describing the effectiveness of pharmacotherapy, which suggest that local pharmacotherapy in SRUS does not give good results, but there are no prospective studies evaluating this method. Various drugs were used in the form of rectal enemas: mesalazine, sulphalazine, steroids, streptokinase, sucralfate, antibiotics, and fibrin glue applied locally.

The exclusion of the rectum from the passage gave the expected results in the SRUS recovery and continu-

ity of the alimentary duct was recreated after a year. Nevertheless, the positive result lasted only for approximately 10 months. The ulceration appeared again, causing small bleeding in the form of mucous slightly coloured with blood. A few therapeutic options, positive experience from the year 1994, and the results of the study by Stoppino *et al*. [6] encouraged the authors to return to argon therapy, but with different objectives. The method of local application was modified by extending the periods between argon coagulations by a month. The expectations and treatment aim were also changed, from stopping the bleeding to healing the ulceration completely. Fifteen APC sessions in one month intervals were done, obtaining a continuous decrease of the large ulceration and as a result, and the full recovery of the SRUS patient after 20 months of therapy. The patient has been under constant clinical control, and no disorders or SRUS relapse have been found for 6 years.

Conclusions

Argon plasma coagulation is a technique that has been used in surgery for over 20 years. The coagulated surface undergoes the processes of superficial coagulation necrosis as a result of mucous membrane blanching, water evaporation and tissue shrinking. This involves only the superficial layer [6, 9, 10]. The APC was effective in stopping the bleeding in our case and other authors' cases. It seems to be effective also in healing solitary rectal ulcer syndrome [6, 10].

The APC therapy in treating SRUS without rectum prolapse could become a crucial method of bleeding treatment in solitary ulcers and an important method (basic and complementary) among the means of SRUS treatment. Temporary exclusion of the rectum from the passage could also be one of the therapeutic options.

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