

Anismo e toxina botulínica: Um caso clínico de sucesso

Anismus and botulinum toxin: A successful clinical case

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RESUMO

O anismo consiste em ausência de relaxamento ou contração paradoxal do puborectalis, durante a defecação. Caso clínico: Mulher de 43 anos, com anismo refratário ao tratamento de reabilitação, tendo-se administrado Toxina Botulínica tipo-A (30 U) no puborectalis. Por persistência de obstipação, efetuámos nova administração (100 U) 4.5 meses depois, verificando-se melhoria subjetiva e objetiva. Repetiu infiltração passados 4.5 meses, apesar de estabilização clínica, que se prolongou por 7 meses adicionais. Realizámos quarta infiltração (100 U) após 8 meses, constatando-se melhoria sintomática acrescida, com três episódios de defecação espontânea. A resolução completa veio após 5ª infiltração, permanecendo assintomática por 28 meses adicionais, data da última avaliação.

Conclusão: A Toxina Botulínica tipo-A revelou eficácia no tratamento do anismo desta paciente, evitando cirurgia e eventuais sequelas permanentes.

ABSTRACT

Anismus refers to a failure of relaxation or paradoxical puborectalis contraction during attempted defecation. Case Report: Anismus in a 43 year-old woman, refractory to rehabilitation treatment. Puborectalis Botulinum Toxin type-A (30 U) injection was done and repeated 4.5 months later (100 U) due to evacuatory difficulty persistence. There was a significant subjective and objective improvement after the second administration, although Botulinum toxin injection (100 U) was repeated 4.5 months later. The fourth BTX-A (100 U) injection took place 8 months after, with symptomatic improvement, referring 3 episodes of spontaneous defecation. Complete resolution came out with the 5th injection, remaining asymptomatic for 28 additional months, dating the last evaluation.

Conclusion: BTX-A was effective in anismus treatment, avoiding surgical decision and its permanent sequelae risks.

INTRODUCTION

Anismus is a functional disorder of evacuation, caused by failed relaxation and/or inappropriate contraction of the striated external anal sphincter during attempted defecation.¹ Its incidence in general population is unknown, being reported in a chronic constipation series, to range from 20% to 70%. It is considered to be more common in women and in young or middle aged individuals.¹ There is a cluster of associated symptoms including prolonged repeated straining, incomplete evacuatory sensation, rectal pain, need for manual disimpaction, laxative or enema abuse.²

Its diagnosis is based on both physical examination and anorectal physiologic testing. An anal sphincter hypertony, a mobility of the posterior loop of the puborectalis during squeezing, as well as the presence of puborectalis squeezing or intermittent contractions when asking to strain, suggests paradoxical puborectalis syndrome.¹ Diagnosis is then confirmed by defecography, manometry, electromyography or dynamic pelvic magnetic resonance imaging.^{2,3} Unfortunately, no single test has been conclusive in determining the presence of anismus.³ Both electromyography and cinedefecography have similar sensitivities and positive predictive values of 67-70%, with specificities hovering around 81%.²

Anismus treatment is highly variable, ranging from biofeedback, botulinum toxin, division of the puborectalis muscle, colectomy, end ileostomy or colostomy.² The primary treatment option is biofeedback, with bowel management reported to have success rates ranging from 40-90%.⁴

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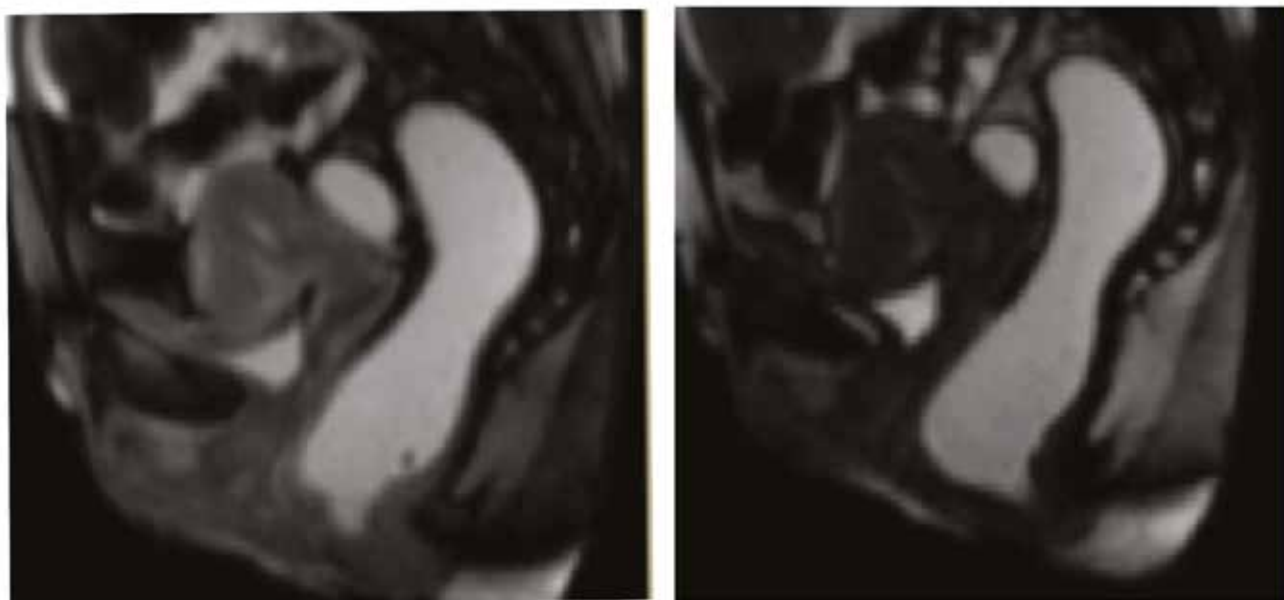


FIGURE 1 Dynamic pelvic magnetic resonance imaging performed five weeks after the second Botulinum Toxin type-A injection. Anorectal angulation at the relaxing position of 108° (left) and during puborectalis voluntary contraction of 102° (right).

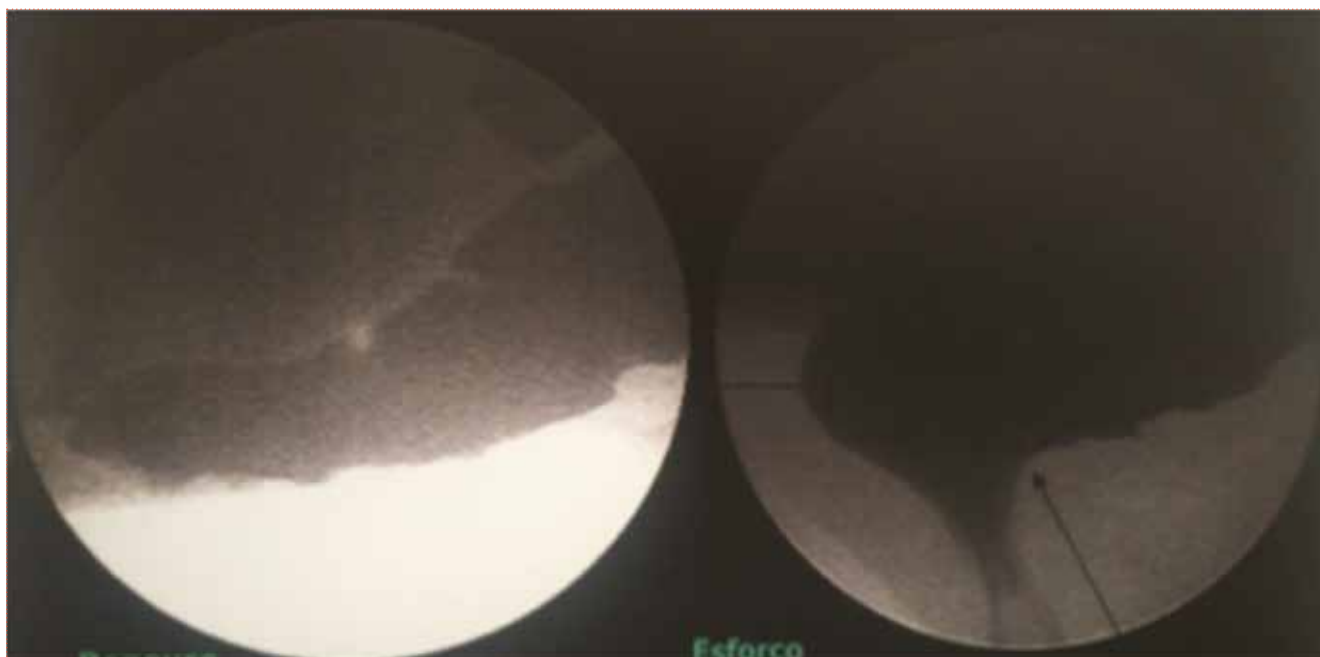


FIGURE 2 Cinedefecography performed 7 months after the 3rd Botulinum Toxin type-A injection, showing failure of puborectalis relaxation during attempted defecation (right).

If patients fail or refuse biofeedback, they are generally then offered neurotoxin *Clostridium botulinum* type A (BTX-A) puborectalis muscle injection, considered extremely successful for temporary treatment of anismus, that is refractory to biofeedback treatment.⁵

CASE REPORT

A 43 year-old female patient, with chronic constipation history since adolescence, with progressive worsening (having endometriosis as the only co-morbidity).

Started several laxatives, with progressive inefficacy, finally doing her bowel emptying through digital stimula-

TABLE 1 Symptom scores and diagnostic test results through time

BTX-A Injection	1 st 13.07.2012		2 nd 30.11.2012		3 rd 22.03.2013		4 th 28.11.2013		5 th 04.07.2014		
Timing of evaluation	Before BTX-A	1.5M after 30 U BTX-A (1 st inj)	1M after 100 U BTX-A (2 nd inj)	3M after 100 U BTX-A (2 nd inj)	2M after 100 U BTX-A (3 rd inj)	7M after 100 U BTX-A (3 rd inj)	1.5M after 100 U BTX-A (4 th inj)	8M after 100 U BTX-A (4 th inj)	6M after 100 U BTX-A (5 th inj)	13M after 100 U BTX-A (5 th inj)	28M after 100 U BTX-A (5 th inj)
Contipation Scoring Scale (0-30)											
	23	23	18	18	12	15	13	9	9	9	9
Visual Analogue Scale (VAS) (0-10)											
VAS (Pain)	10	10	7	7	1	1	1	1	0	1	1
VAS (personal dissatisfaction)	10	10	4	4	2	2	2	1	1	1	1
Dynamic Pelvic MRI and Cinedefecography											
Anorectal angle rest/ attempted defecation	110°/65°		108°/102°			95°/115°					100°/95°
Complete rectal evacuation	No		No			No					No
Anorectal Monometry											
Anal pressure rest / attempted defecation	80/130	95/58	76/31	90/100	99/71	102/58	91/35				88/60
Anal sphincter pressure during attempted defecation	↑	↓	↓	↓	↓	↓	↓				↓
Ballon expulsion test	NP		P+T / P 50 mL - 4" 100 mL - 0"	P+T 4,6 50 mL - 4" 100 mL - 6"	P+T 50 mL - 5" 100 mL - 2"	P+T 50 mL - 5" 100 mL - 4"	P				P+T 50 mL - 5" 100 mL - 4"
*BTX - A - Botulinum toxin type-A; ↑ increased; ↓ decreased; NP not possible; P+T Possible with manual traction (time of traction for 50 and 100mL ballon); P Possible without manual traction.											

tion, associated with water glycerine enema application.

Anismus was only diagnosed at 39 years-old through anamnesis, magnetic resonance defecography and anorectal manometry. The first imaging examination

revealed anorectal narrowing angulation, from relaxed position to defecation attempt (from 110° to 65°), with puborectalis paradoxical contraction and exerting pressure over the posterior rectum wall, as well as totally in-

complete defecation. Anorectal manometry revealed anal sphincter pressure increment during defecation attempt, compatible with dyssynergic defecation, with balloon expulsion test totally ineffective.

Patient had done a 3 year rehabilitation program, including biofeedback, endoanal electrical stimulation, acupuncture, osteopathy, without any clinical improvement. She then went to a Physical Medicine and Rehabilitation consultation, having been decided for a rehabilitation plan twice weekly, including muscle anal sphincter and pelvic floor relaxation massage, hypopressive abdominal exercises (both supervised and home exercise programme, daily). She was prescribed diazepam (5 mg per day) and domperidone (10 mg, 3 times per day).

Although abdominal pain improved, a defecation difficulty persisted Scoring Constipation System Scale (CSS) and Visual Analogic Scale (VAS) for "pain" and "personal dissatisfaction", having been decided for the first BTX-A puborectalis infiltration (30 U). This procedure was done without sedation and in the supine position, in gynecology table (contrary to the literature, which describes the technique as being done in left lateral position).

A 0.30 mL volume (of 1 mL containing 100 U Botox®, Allergan), corresponding to 30 U of toxin was injected into the left and right sides of the puborectalis and external anal sphincter at 5 and 7 o'clock position (15 U in each side). A syringe fitted to a needle size of 21G and 50 mm length was used, being the needle tip guided by the left index finger into the anal canal.

Due to constipation and abdominal pain persistence 1.5 months after toxin injection, with subjective and objective parameters equal to the pre-toxin state, despite improvement in manometric parameters (with anal pressure reduction during defecation attempt), it was decided for the 2nd BTX-A injection (100U, with 50U in each side of the anal sphincter) 4.5 months after the first one.

Subjective and objective improvement was noticed 1 month after the last procedure, comparing both to the pre-toxin and to the 30U post-toxin states. Manometrically, there was a stabilization, with anal pressure reduction during defecation attempt, as well

as an effective balloon expulsion test.

A magnetic resonance defecography was done in the same period, showing clinical improvement comparing to the pre-toxin state, with a constant ano-rectal angulation during defecation, resting position or during puborectalis voluntary contraction (of 102°, 108° and 100°, respectively), although still with a moderate puborectalis impression over the posterior ano-rectum wall (Figure 1).

Due to patient fear of clinical worsening, although with manometric and scoring for symptoms/signs stabilization, a third toxin injection was done (50 U+50 U), 4.5 months after the second one (Figure 2).

A new clinical worsening came out seven months later, with low caliber stool and anal hypertony at manometry during sphincter relaxation attempt, despite ano-rectal angulation improvement (from 95° in relaxing position to 115° during defecation attempt), being decided for the 4th injection (100U), 8 months after the previous one. Patient then referred a continuous subjective improvement since the beginning of the toxin treatment (CSS changing from 23 in the pre toxin state to 13 1.5 months after this last procedure, as well as changing in VAS scale, from 10 to 1 respectively, concerning both pain and personal dissatisfaction), describing 3 episodes of spontaneous defecation with only digital stimulation. Complete clinical resolution came out after the 5th toxin infiltration, 8 months after the previous one, in spite of soft abdominal pain reappearance (VAS 2/10) (Table 1).

The last evaluation, occurring 28 months after the last injection, revealed worsening on cinedefecography (although the patient felt the exam didn't correspond to her usual bowel habits). However, manometric results improved, with complete symptomatic resolution and spontaneous diary defecation. (CSS of 9 ; VAS of 1, for both pain and personal dissatisfaction).

CONCLUSION

BTX-A revealed efficacy in anismus treatment of this particular patient, with apparent cumulative effect. Although multiple injections were performed, its necessity was spread in time, reaching a definite and successful treatment interruption.

The authors present this case report because of its



scientific innovation and positive results obtained, avoiding aggressive surgical intervention with permanent sequelae risks. ●●●

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