DOI: 10.4274/tjcd.28863
Turk J Colorectal Dis 2018;28:22-26

RESEARCH ARTICLE

Anal Bowen's Disease: Retrospective Analysis of Five Cases

Anal Bowen Hastalığı: Beş Olgunun Retrospektif Analizi

Ozan Akıncı, Fadime Kutluk, Süphan Ertürk, Serdar Yüceyar
İstanbul University Cerrahpaşa Faculty of Medicine, Department of General Surgery, İstanbul, Turkey

ABSTRACT

Aim: Anal Bowen's disease is a rare *in-situ* squamous cell carcinoma of the anus. Symptoms such as pruritus, burning, and pain in the perianal region are the first complaints of patients at admission. The disease should be differentiated from many dermatological diseases such as psoriasis, Paget's disease, eczema, seborrheic dermatitis, and lichen planus. Factors such as human papilloma virus, arsenic, radiation exposure, and immunosuppression have been idefined in the etiology. In this study, we retrospectively reviewed five patients who were diagnosed with and treated for anal Bowen's disease in our clinic.

Method: We retrospectively analyzed parameters such as age, gender, symptoms, lesion size, surgical treatment technique, comorbidity, and predisposing factors of five patients who were diagnosed with anal Bowen's disease and treated surgically in our clinic between 2000 and 2017.

Results: The mean age of the cases was 64 years. Three were female and two were male. The most common symptom was anal itching. One of the patients refused treatment, while two underwent excision + primary suture. One patient underwent excision + Limberg flap, while excision + V-Y advancement flap were performed in the other case.

Conclusion: Anal Bowen's disease is a rare *in-situ* squamous cell carcinoma that is diagnosed by lesion biopsy. Wide surgical excision is a safe and definitive treatment. Primary repair or skin flaps can be used for reconstruction.

Keywords: Anal Bowen's disease, surgical, treatment

ÖZ

Amaç: Anal Bowen hastalığı anüsün nadir görülen *in-situ* skuamöz hücreli kanseridir. Perianal bölgede kaşıntı, yanma, ağrı gibi semptomlar hastaların ilk başvuru sebepleridir. Hastalığın psöriazis, Paget hastalığı, egzema, seboreik dermatit, liken planus gibi birçok dermatolojik hastalıkla ayrımı yapılmalıdır. Etiyolojide arsenik, radyasyon maruziyeti, human papilloma virüs, immünosüpresyon gibi faktörler tanımlanmıştır. Bu yazımızda kliniğimizde teşhisi konularak tedavi edilen beş anal Bowen olgusunu retrospektif olarak inceledik.

Yöntem: Kliniğimizde 2000-2017 yılları arasında anal Bowen hastalığı tanısı konulan ve tedavi edilen beş olguda yaş, cinsiyet, semptom, lezyon boyutu, cerrahi tedavi tekniği, eşlik eden hastalık ve predispozan faktörler retrospektif olarak incelendi.

Bulgular: Olguların yaş ortalaması 64, üçü kadın ikisi erkek olup en sık görülen semptom anal kaşıntıydı. Olgulardan biri tedaviyi reddetmiş, ikisine eksizyon + primer sütür uygulanmıştır. Bir olguda eksizyon + Limberg flebi diğerinde ise V-Y ilerletme flebi uygulanmıştır.

Sonuç: Anal Bowen hastalığı nadir görülen *in-situ* skuamöz hücreli karsinomdur. Tanısı lezyondan biyopsi ile konulur. Tedavisinde geniş cerrahi eksizyon kür sağlayıcı ve güvenilir bir yöntem olup rekonstruksiyon için primer tamir veya deri flepleri uygulanabilir.

Anahtar Kelimeler: Anal Bowen hastalığı, cerrahi, tedavi

Introduction

Perianal tumors are rare pathologies and they make up 3-4% of anorectal cancers.[1,2] Bowen's disease was firstly described by John T. Bowen in 1912 as a premalignant lesion occurring on the face, neck, arms, trunk, and perianal region. Bowen's disease is defined as keratinized or non-keratinized intra-epithelial squamous cell carcinoma (SCC) of the anal region. It is also described as the high-grade form of anal intraepithelial neoplasia.[3,4] It is characterized by erythematous, well-defined, brown-red pigmented plaques or nodules on the perianal skin. It most commonly occur

Address for Correspondence/Yazışma Adresi: Ozan Akıncı MD
İstanbul University Cerrahpaşa Faculty of Medicine, Department of General Surgery, İstanbul, Turkey
Phone: +90 537 749 06 41 **E-mail:** ozanakinci1987@hotmail.com **ORCID ID:** orcid.org/0000-0002-7149-6854
Received/Geliş Tarihi: 03.01.2018 **Accepted/Kabul Tarihi:** 11.02.2018

between the ages of 50-80 years, and equally in males and females.[5,6] The most important factor in the etiology is human papilloma virus (HPV) infection (especially type 16 and 18).[5,7] The definite diagnosis is made with biopsy. Since it is a pre-cancerous lesion, the curative treatment is surgical excision. When surgery is not appropriate or the patient refuses surgical treatment, alternative treatment methods (cryotherapy, photodynamic therapy, radiotherapy, 5-fluorouracil, imiquimod, etc.) can be administered.

Materials and Methods

The parameters such as age, gender, symptoms, lesion size, surgical treatment technique, comorbidity and predisposing factors of five cases who were diagnosed with anal Bowen's disease and treated surgically in our clinic between the years of 2000-2017 were examined retrospectively (Table 1). The information regarding the final status of the patient was obtained by making phone interview with the subjects. Four cases were treated surgically while one of them refused treatment. Consent form was filled out by all participants.

Results

In all cases, the lesions were well-defined, dry-looking, erythematous plaques macroscopically (Figure 1, 2). The mean age of the cases was 64 years. Of them, three were female and two were male. The most common symptom was anal itching. One of the cases (case 3) refused the treatment, two of them (case 1, 2) underwent excision + primary suture. In one case (case 4), excision + Limberg flap was performed while excision + V-Y advancement flap were performed in the other case (case 5). Predisposing factor was determined to be HPV in one case while it was found to be immunosuppressive treatment in one case who received it due to systemic lupus eritemotozis (SLE) with renal involvement. One of the cases had also SCC on the scalp. Two cases in whom primary suture was performed were lost to follow-up. No complication associated with surgical treatment developed in the case in whom Limberg flap was used.

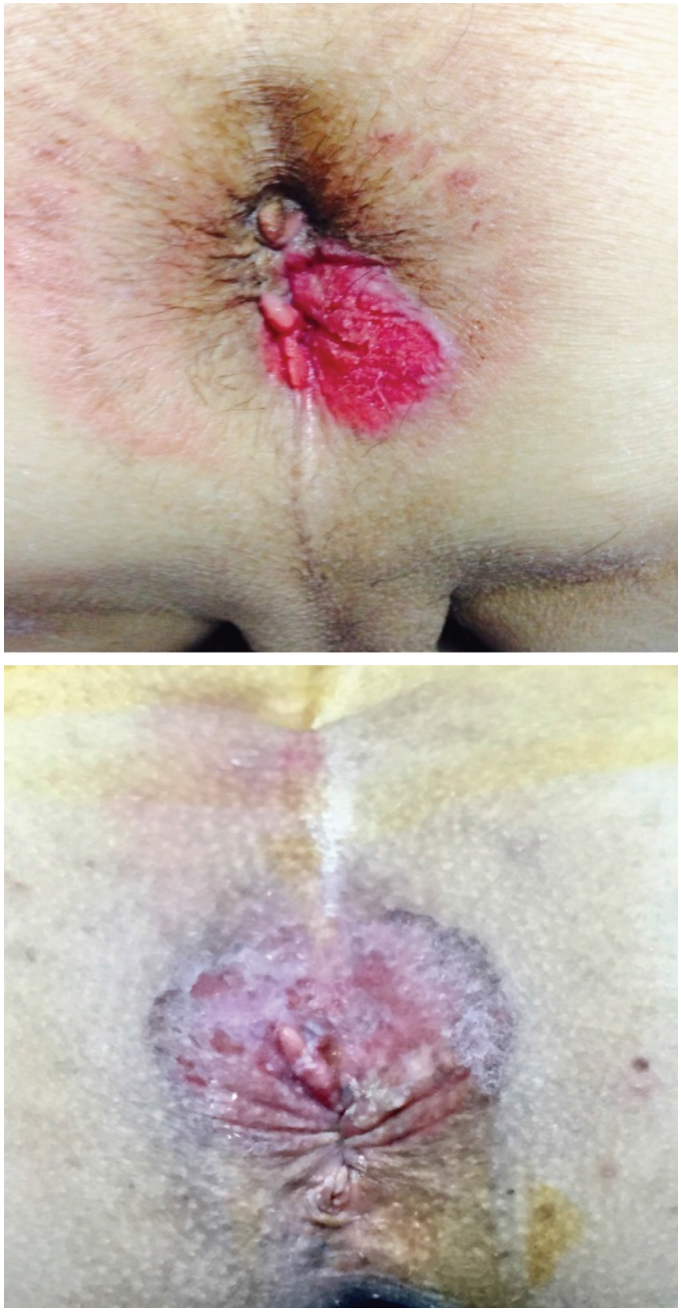

Figure 1, 2. The appearance of Bowen's disease in case 4 and 5

Table 1. The characteristics and treatments of the cases

Age of patient	Gender	Symptom	Size of lesion (cm)	Surgical treatment	Predisposing factor	Comorbidity
1-81	F	Itching	0.7*0.3	Excision + primary suture	-	-
2-79	M	Itching, pain	1*0.5	Excision + primary suture	-	SCC (on the scalp)
3-42.	F	Itching, palpable nodüle	0.4*0.7	Refusal of treatment	HPV	-
4-85	M	Itching	3.5*3.3	Excision + Limberg flap	-	-
5-37	F	Itching, redness	4.5*2.5	Excision + V-Y advancement flap	Immunosuppressant treatment	SLE

F: Female, M: Male, HPV: Human papilloma virus, SCC: Squamous cell carcinoma, SLE: Systemic lupus eritemotozis

The lesion of the case, in whom V-Y advancement flap was performed (Figure 3, 4, 5), was extending over the mucosa of anal canal, adjacent to dentate line. Temporary gas and fluid incontinence was detected in the early postoperative period due to the wide excision of the mucosa along with the perianal skin, however it recovered within two months spontaneously. Since the biopsy result of the same case revealed that involvement continued as islets, the case was operated on again after five months. Re-excision so that the previous incision lines would be removed and one cm of macroscopically intact borders would be attained + repair with V-Y advancement flap from three quarters were performed (Figure 6). Since the biopsy result was positive

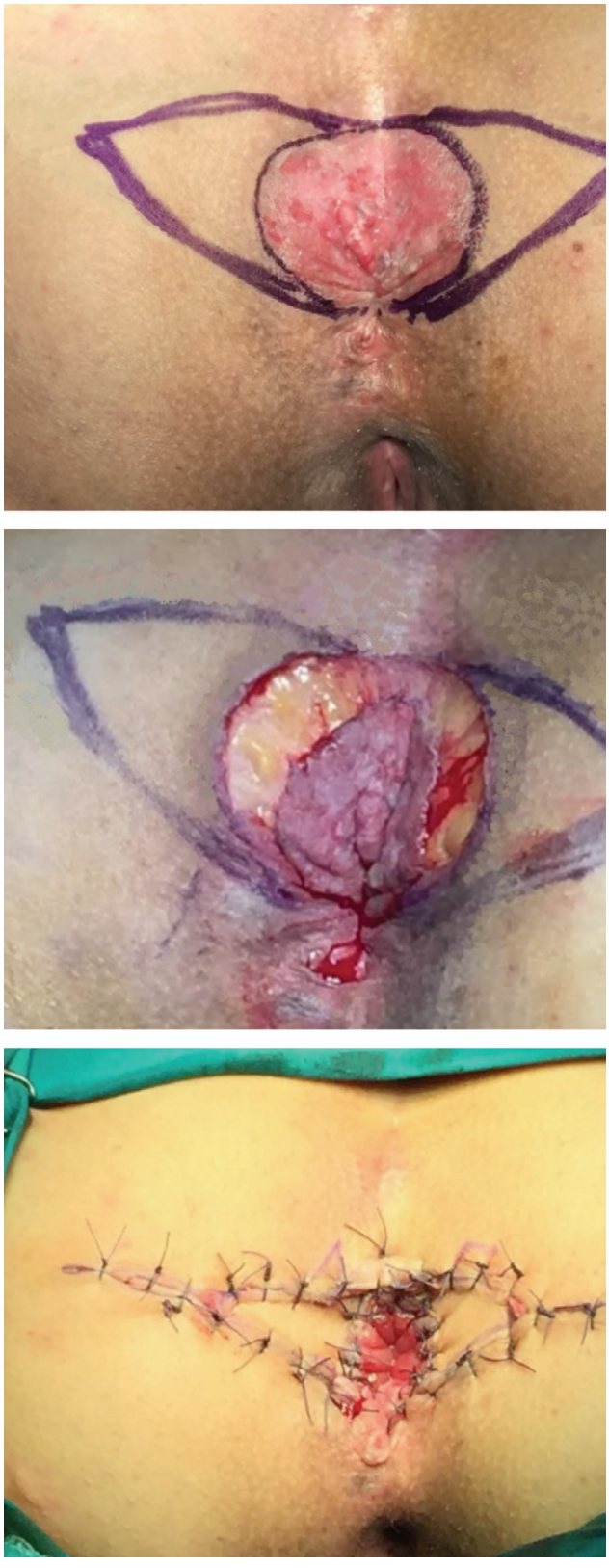

Figure 3, 4, 5. Excision and repair with V-Y advancement flap in case 5

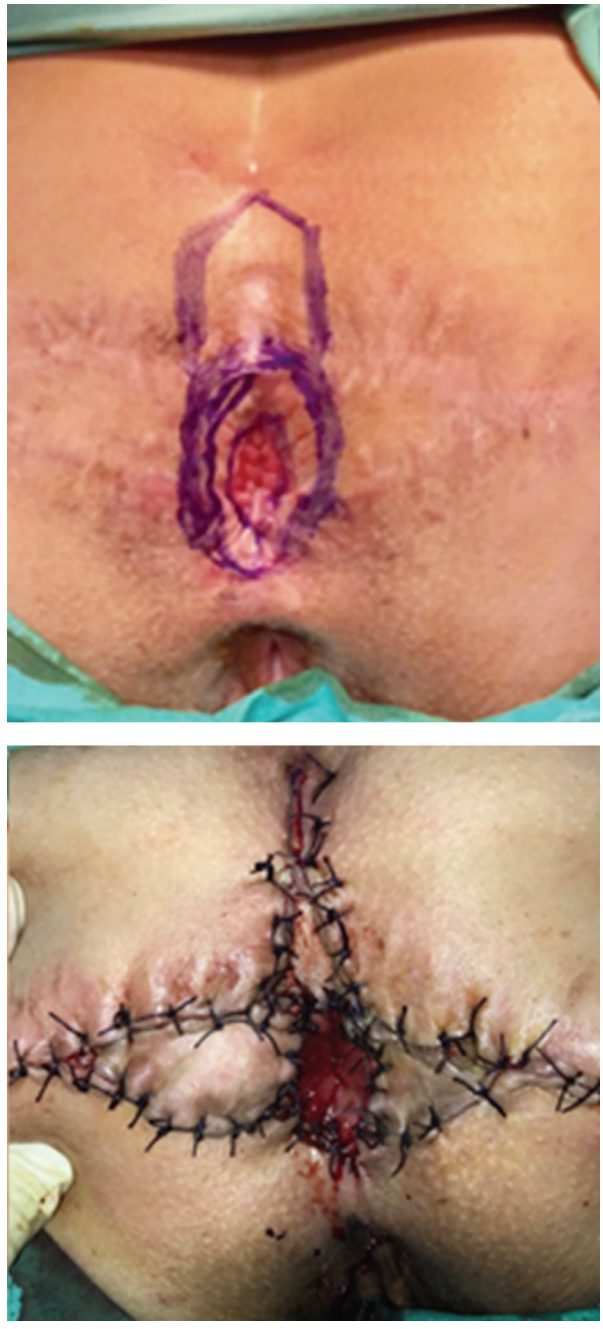

Figure 6. Re-excision and repair with triple V-Y advancement flap in case 5

again, a new excision was not performed with the concern that it could create problems of anal functions. Due to the presence of SLE, radiotherapy was considered risky and local imiquimod administration by dermatologists was planned.

Discussion

Bowen's disease is an in-situ SCC which appear on the face, neck, trunk, arms, genitalia, and peri-anal region and rarely invades adjacent organs or metastases (5%).[8] The likelihood of transformation to invasive SCC is 2-6%.[9] Anal localization is seen quite rarely in Bowen's disease. It is characterized by erythematous, well-defined, brown-red pigmented plaques or nodules on the perianal skin. Dermatological diseases such as Paget's disease, psoriasis, nummural dermatitis, seborrheic dermatitis, lichen planus, lupus vulgaris, secondary syphilis should be considered in the differential diagnosis.[5,6,7] It may present with symptoms such as anal itching, pain and palpable nodules. Forty percent of the cases are asymptomatic and they are often detected incidentally in hemorrhoidectomy specimens.[9] All five cases in our study were symptomatic and the most common symptom was itching. HPV is most commonly responsible for the etiology, and immunosuppression, arsenic and radiation exposure are also identified as other risk factors.[5,7,10] HPV infection in one of our cases and immunosuppressive treatment in another case were determined as the predisposing factors. The definite diagnosis of Bowen's disease is made by histopathological examination of biopsy material. Since it is a premalignant lesion, the curative standard treatment is wide surgical excision.[6] Some authors recommended a four-quadrant biopsy technique (mapping technique) in which multiple frozen sections were performed intraoperatively in order to get negative surgical margins.[7] These frozen sections should also include the intra-anal mucosal biopsy. The mapping technique was reported to be an effective way of determining the width of lesion,[11] but despite this technique, the recurrence rate was reported to be up to 30%.[7,12] In a retrospective clinical trial including 617 cases, surgical excision was shown to be one of the treatments with the lowest relapse rate at 5 years follow-up.[13] The major disadvantages of wide excision are that it is difficult to close the defect primarily and that it may require skin flap. For the reconstruction of defect, primary repair, secondary healing, gradual excision combined with skin flap, rotation flap, myocutaneous flap and V-Y advancement flaps can be used. Complications such as flap necrosis, anal stenosis, incontinence and sexual dysfunction may occur after surgical excision and reconstruction. Temporary gas and fluid incontinence developed in one of our cases in whom V-Y advancement flap was used. Less aggressive treatment modalities were tried since problems associated with wide skin defects after wide surgical excision and transformation of the lesions to SCC happened relatively less commonly. These methods include cryotherapy, argon laser therapy, photodynamic therapy, radiotherapy, 5-fluorouracil, infrared coagulation, imiquimod and close follow-up.[14,15,16,17,18,19] Especially, recent treatment guidelines report that radiotherapy is also highly effective and safe.[6]

There is no standard protocol for follow-up of anal Bowen's disease. Beck recommends annual physical examination, flexible sigmoidoscopy, also punch biopsy and random biopsy from the margins of the skin grafts if new lesion emerges.[20] Relapse was reported nine years after wide surgical excision in the literature.[4] Therefore, follow-up period may need to encompass a period of time longer than 5 years. Anal Bowen's disease is a rarely occurring in-situ SCC. It is diagnosed with the biopsy taken from the lesion. Wide surgical excision is a safe and curative method for the treatment. Primary repair or skin flaps can be used for reconstruction. Due to the high rate of relapse, follow-up should be continued in the long term.

Ethics

Ethics Committee Approval: Retrospective study.

Informed Consent: Consent form was filled out by all participants.

Peer-review: Internally peer-reviewed.

Authorship Contributions

Surgical and Medical Practices: S.E., S.Y., O.A., Concept: O.A., S.E., Design: O.A., F.K., Data Collection or Processing: O.A., F.K., Analysis or Interpretation: O.A., F.K., Literature Search: O.A., F.K., Writing: O.A.,

Conflict of Interest: No conflict of interest was declared by the authors.

Financial Disclosure: The authors declared that this study received no financial support.

References

1. Strauss RJ, Procaccino JA, Moffa MA. Bowen's disease and Paget's disease. In: Fazio VW, Church JM, Delaney CP, eds. Current Therapy in Colon and Rectal Surgery. 2nd ed. Philadelphia: ElsevierMosby; 2005:75-78.
2. Gervasoni JE Jr, Wanebo HJ. Cancers of the anal canal and anal margin. Cancer Invest 2003;21:452-464.
3. Skinner PP, Ogunbiyi OA, Scholefield JH, Start RD, Smith JH, Sharp F, Rogers K. Skin appendage involvement in anal intraepithelial neoplasia. Br J Surg 1997;84:675-678.
4. Northfett DW, Swift PS, Palefsky JM. Anal neoplasia. Pathogenesis, diagnosis, and management. Hematol Oncol Clin North Am 1996;10:1177-1187.
5. Cleary RK, Schaldenbrand JD, Fowler JJ, Schuler JM, Lampman RM. Perianal Bowen's disease and anal intraepithelial neoplasia: review of the literature. Dis Colon Rectum 1999;42:945-951.

6. Cox NH, Eedy DJ, Morton CA. Therapy Guidelines and Audit Subcommittee, British Association of Dermatologists. Guidelines for management of Bowen's disease: 2006 update. Br J Dermatol 2007;156:11-21.
7. Marchesa P, Fazio VW, Oliart S, Goldblum JR, Lavery IC. Perianal Bowen's disease: a clinico pathologic study of 47 patients. Dis Colon Rectum 1997;40:1286-1293.
8. Heidi Nelson. Anus. Chapter 53, In Sabiston Textbook of Surgery, 19th ed. Elsevier, Philadelphia, 2012.
9. Thaler K, Wexner SD. Cancers of the Anal Margin and Anal Canal, Bowen's Disease, and Paget's Disease. Operative Techniques in General Surgery 2001;3:171-182.
10. Margenthaler JA, Dietz DW, Mutch MG, Birnbaum EH, Kodner IJ, Fleshman JW. Outcomes, risk of other malignancies, and need for formal mapping procedures in patients with perianal Bowen's disease. Dis Colon Rectum 2004;47:1655-1660.
11. Beck DE, Fazio VW, Jagelman DG, Lavery IC. Perianal Bowen's disease. Dis Colon Rectum 1988;31:419-422.
12. Sarmiento JM, Wolff BG, Burgart LJ, Frizelle FA, Ilstrup DM. Perianal Bowen's disease: associated tumors, human papilloma virus, surgery, and other controversies. Dis Colon Rectum 1997;40:912-918.
13. Thestrup-Pedersen K, Ravnborg L, Reymann F. Morbus. Bowen. A description of the disease in 617 patients. ActaDerm. Venereol 1988;68:236-239.
14. Boynton KK, Bjorkman DJ. Argon laser therapy for perianal Bowen's disease: a case report. Lasers Surg Med 1991;11:385-387.
15. Jones CM, Mang T, Cooper M, Wilson BD, Stoll HL. Photodynamic therapy in the treatment of Bowen'sdisease. J Am Acad Dermatol 1992;27:979-982.
16. Runfola MA, Weber TK, Rodriguez-Bigas MA, Dougherty TJ, Petrelli NJ. Photodynamic therapy for residual neoplasms of the perianal skin. Dis Colon Rectum 2000;43:499-502.
17. Ramos R, Salinas H, Tucker L. Conservative approachto the treatment of Bowen's disease of the anus. Dis Colon Rectum 1983;26:712-715.
18. Goldstone SE, Kawalkek AZ, Huyett JW. Infrared coagulator: a useful tool for treating anal squamous intraepithelial lesions. Dis Colon Rectum 2005;48:1042-1054.
19. Moreno G et al. Therapeutic options for Bowen's disease. Austral J Derm 2007;48:1-10.
20. Beck DE. Paget's disease and Bowen's disease of the anus. Semin Colon Rectal Surg 1995;6:143-149.