

Preservation of Anal Function After Total Excision of the Anal Mucosa for Bowen's Disease

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Six women with Bowen's disease of the anogenital area were treated by total excision of the anal mucosa, perianal skin and, in some cases, partial vulvectomy. Two patients had foci of microinvasive squamous carcinoma. Adequate tumor margins were determined by frozen sections. The resulting mucosal and cutaneous defects were grafted with medium split-thickness skin grafts applied to the anal canal and sutured circumferentially to the rectal mucosa. Grafts were held in place by a finger cot inserted in the anal canal and stuffed with cotton balls. Patients were constipated five or six days with codeine. The skin grafts healed per primam. One additional patient was similarly treated for a chronic herpetic ulceration of the anus and healed. Contrary to dire predictions, all patients were able to distinguish between gaseous and solid rectal contents and sphincter function was preserved. In one patient, Bowen's disease has recurred in the grafted perianal skin.

INTRAEPITHELIAL CARCINOMA (BOWEN'S DISEASE) of the anus is being seen with increasing frequency in association with *in situ* carcinoma of the vulva and is being detected in younger women than in the past.^{1,2} The reason for this apparent increase in frequency is unknown. Infection by herpes simplex virus type 2 and the genital wart virus are prime suspects in carcinomas arising in the female lower genital tract.³ The extensive involvement of the anal mucosa by Bowen's disease and its potential to become invasive carcinoma have necessitated an innovative surgical approach that is concerned with cure as well as preservation of function of the anus. The current study reviews the authors' experience with this problem and describes our method of surgical management.

Methods

During the 4-year period from August 1978 to August 1982, six patients were treated for carcinoma *in situ* of the anal mucosa and perianal skin (Fig. 1). Two of these patients had microinvasive squamous carcinoma in the involved skin or mucosa (Fig. 2). Four patients had syn-

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chronous carcinoma *in situ* of the vulva and two patients had metachronous carcinoma *in situ* of the vulva. One patient with *in situ* carcinoma of the vulva had an area of minimally invasive squamous carcinoma. Four patients gave a history of having had condyloma acuminatum of the perianal area or vulva treated in the past. In one patient there was a history of genital herpes infection. These six cases are summarized in Table 1.

All biopsy specimens were reviewed and the diagnosis of squamous cell carcinoma *in situ* and microinvasive squamous carcinoma confirmed. Before surgery, patients were screened with Pap smears of the cervix and vagina and colposcopy. Biopsies of the cervix, vulva, and perianal skin were taken where appropriate (Fig. 3). Proctoscopy was performed on each patient. Preoperative biopsies of vulva and anal mucosa were occasionally taken under anesthesia to determine the extent of the disease.

All patients had total excision of the anal mucosa and involved perianal skin and four of these were combined with a partial vulvectomy or total skinning vulvectomy done in continuity with the anal excision. With the patient in the lithotomy position, the skinning vulvectomy is carried out first by the gynecologic team. The dissection is continued around the anus and into the anal canal by the general surgical team. The anal mucosa and submucosa is dissected away from the underlying sphincter and continued inward beyond the dentate line (Fig. 4). The specimen is submitted to the pathologist who checks the surgical margins with multiple frozen sections. Further excision of mucosa above the dentate line well into the transition zone was necessary in three patients before margins free of *in situ* carcinoma were obtained. In two cases this necessitated excision of the mucosa approximately 1.5 cm above the dentate line. Strict attention to hemostasis is essential.

When all margins are declared free of tumor, the plastic surgical team harvests medium split-thickness skin grafts

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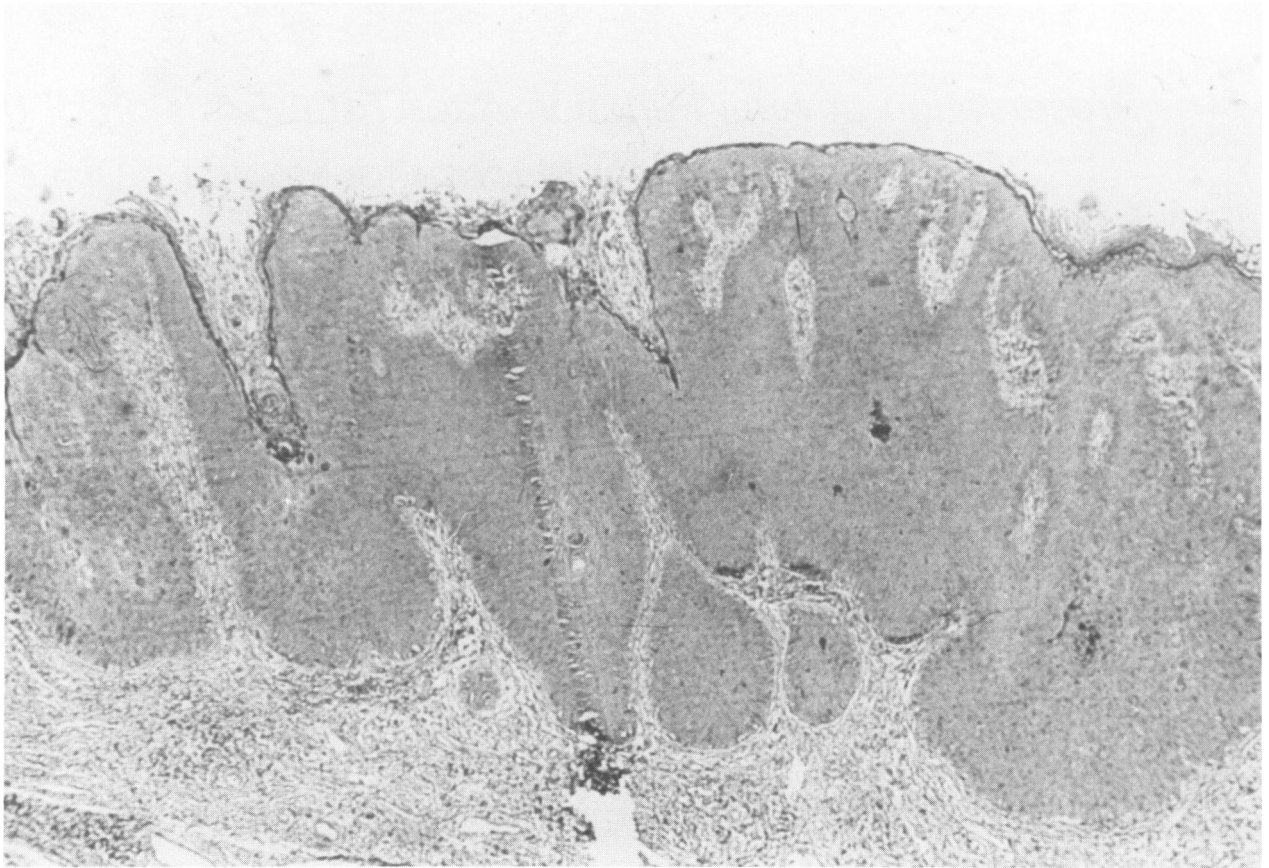


FIG. 1. Microscopic section showing thickened epidermis with grave cytologic atypia characteristic of Bowen's disease.

with a Reese dermatome from the previously prepared buttocks. These grafts are applied in two halves and sutured to the perianal skin margin with long silk ties from the 12:00 to 6:00 position on one side and the 6:00 to 12:00 position on the other. The grafts are sutured to each other and to the rectal mucosa with interrupted sutures of 4-0 Dexon (Fig. 5). Compression of the grafts to the anal canal is maintained by a finger cot filled with cotton balls for bulk and a Xeroform® gauze dressing wrapped and sutured to the finger cot. This obturator is carefully inserted into the anal canal and lower rectum and then a dressing of Xeroform gauze and moist cotton balls is applied to the perianal and vulvar grafts. The stint is then secured by tying the long sutures over the dressings.

Good mechanical bowel preparation with a clear liquid diet, magnesium citrate, castor oil, and saline enemas prior to operation is an essential part of the procedure. Before surgery a single injection of a cephalosporin is given and continued several days after surgery. An indwelling Foley catheter is placed in the bladder at the beginning of the operation and remains until after the dressing is removed. After surgery the patient is kept from defecating 5 or 6 days with codeine or Lomotil. Normal

defecation occurs after removal of the dressing and stool softeners are given during the first few weeks after operation.

Results

The ages of the patients studied ranged from 24 to 42 years, with an average age of 33.7. All six patients were white. Four patients were referred with a diagnosis of carcinoma *in situ* of the vulva and on further evaluation were found to have anal involvement. One of these patients also had minimally invasive squamous carcinoma of the vulva. Two patients had previously undergone a hysterectomy for carcinoma *in situ* of the cervix. One of these women subsequently had a partial vulvectomy for *in situ* carcinoma. She was referred to the Gynecologic Oncology Department for follow-up, and biopsy of a small perianal ulcer disclosed an invasive squamous carcinoma in association with Bowen's disease. A 32-year-old patient had a "hemorrhoid" excised from her anal canal as an office procedure and histologic examination revealed microinvasive squamous carcinoma in an extensive area of Bowen's disease. An abdominoperineal resection was

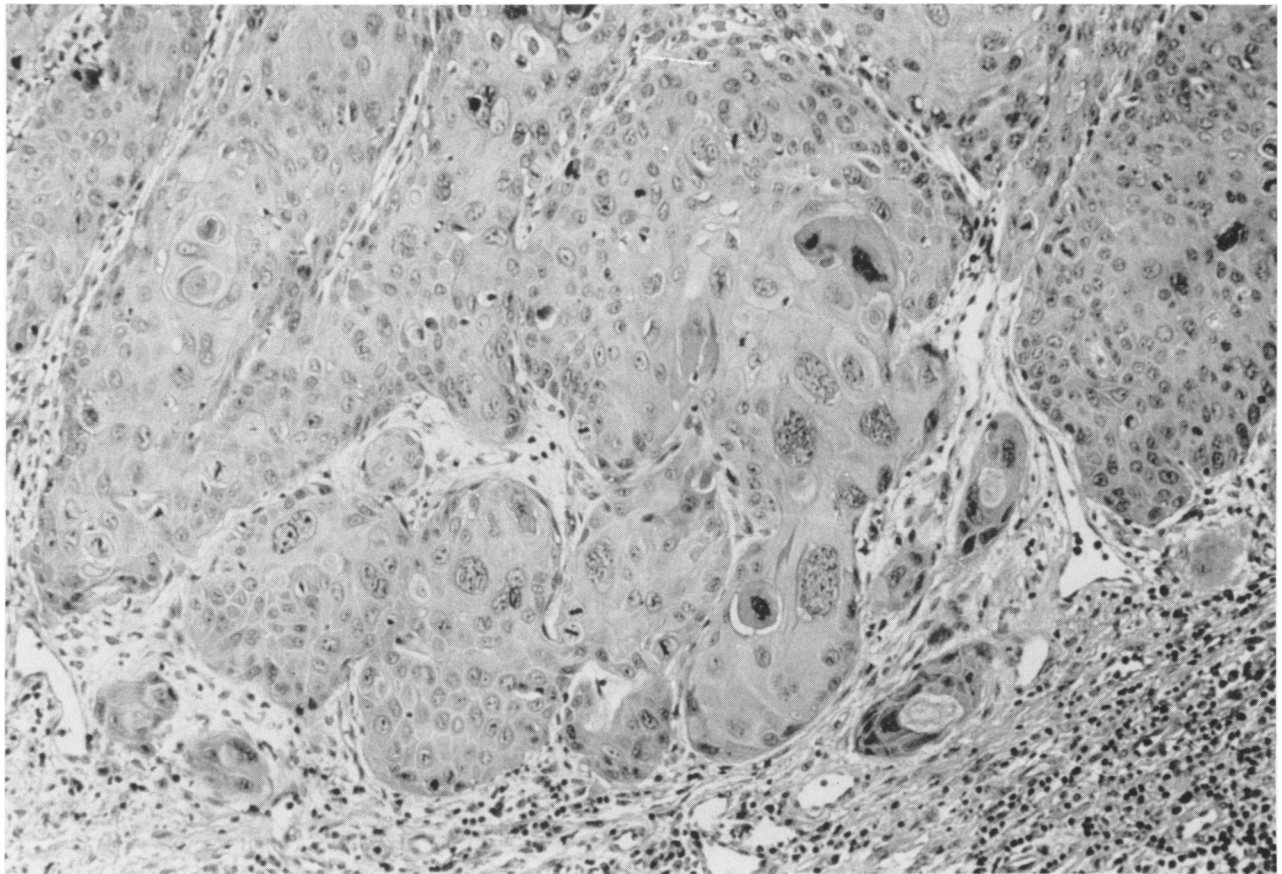


FIG. 2. Microscopic section showing a focus of microinvasive squamous carcinoma in continuity with Bowen's disease.

considered; however, the patient refused this procedure and sought an alternative. This patient and three others had all been treated in the past for perianal or vulvar condyloma acuminatum. One other patient had a history of genital herpes.

Four of the six patients were treated by partial or total skinning vulvectomy in continuity with the involved perianal skin and entire anal mucosa. The resulting defect was covered with split-thickness skin grafts. Two patients had excision of the perianal skin and anal mucosa only.

TABLE 1. Summary of Cases of Carcinoma In Situ of the Anal Canal and Perianal Skin

Age (years)	Race	Other Conditions*	Initial Treatment†	Date	Recurrences and Treatment
36	W	CIS cervix CIS vulva Condyloma	Wide excision	8/78	10/80: Dysplasia anorectal junction; laser Vaporization
42	W	CIS cervix CIS vulva Condyloma	TEAM + STSG PV + TEAM + STSG	11/78 1/79	9/80: Condyloma 2/81: Laser vaporization 8/82: CIS in graft Biopsy only
24	W	CIS vulva	PV + TEAM + STSG	4/80	None
32	W	Dysplasia cervix Condyloma	TEAM + STSG	12/81	None
31	W	CIS vulva Condyloma	SV + TEAM + STSG	5/82	None
37	W	CIS vulva Dysplasia cervix Genital herpes	SV + TEAM + STSG	8/82	None

* CIS—carcinoma *in situ*.

† TEAM—total excision anal mucosa.

STSG—split-thickness skin graft.

PV—partial vulvectomy.

SV—skinning vulvectomy.

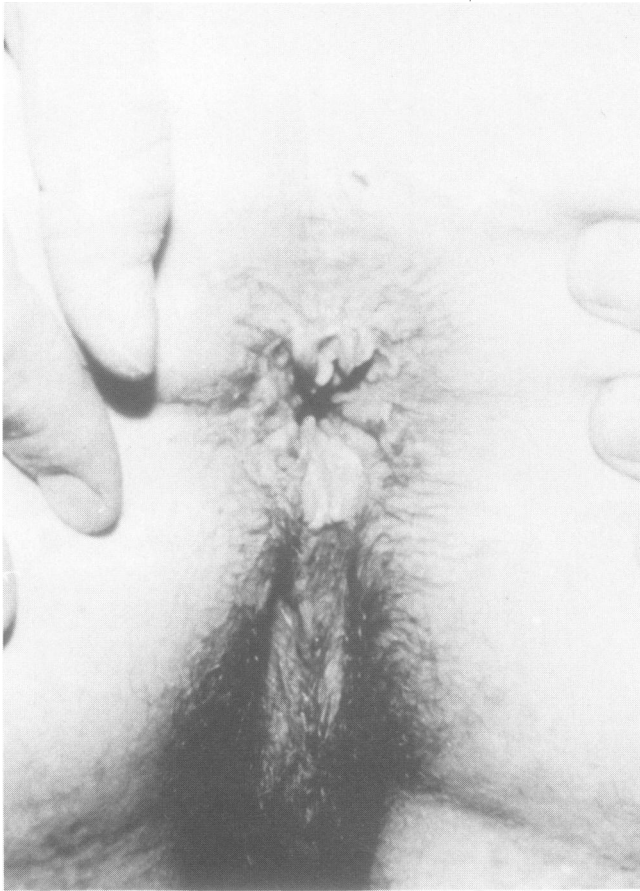


FIG. 3. Multiple perianal skin taga all extensively involved with Bowen's disease.

Skin grafts healed on initial grafting in four patients. Two patients required a second graft to the anal canal in the immediate postoperative period and healed primarily. After removing the dressing, most patients experienced transient fecal soiling that cleared completely in 4 to 6 weeks. Two patients required daily enemas for several weeks to reestablish regular bowel movements. One additional patient with a painful chronic herpetic ulceration of the anus has been treated in a similar fashion and now maintains normal anal function. They are able to distinguish between solid and gaseous contents in their rectum. Sphincter tone is excellent and no anal stenosis has resulted (Fig. 6).

These six patients have now been followed 1 to 5 years. An area of dysplasia at the junction of the graft and the rectal mucosa was discovered in the first patient 2 years after operation and was treated by laser vaporization. There has been no further recurrence. Of particular interest was the development of condyloma acuminatum in the grafted skin of the vulva and perineum in the second patient 2 years after her operation. The condylomata were treated by laser vaporization. One and one-half years after laser vaporization, pigmented plaques ap-

peared in her vulvar and perianal skin graft and biopsy confirmed the recurrence of Bowen's disease. This recurrence is currently being followed without treatment and there has been no progression of the disease. The other four patients are free of disease at the time of this report.

Discussion

Squamous carcinoma *in situ* of the vulva frequently involves the perianal skin and anal mucosa and can be adequately treated by total excision of this intraepithelial carcinoma. In the authors' experience when anal involvement is present, it nearly always involves the anoderm circumferentially up to the dentate line. In three patients tumor was present in the margins of resection and further excision above the dentate line was necessary before margins free of tumor were obtained. The resulting defect presents a considerable surgical problem. If the wound were left to heal secondarily, anal stricture would almost certainly result. Bringing down the rectal mucosa and suturing it to the perianal skin to cover the exposed anal sphincter (Whitehead's operation) yields a very unsatisfactory result according to Goligher.⁵ The exposed rectal mucosa is subjected to constant trauma and bleeds easily. Mucus production causes unacceptable soiling, maceration of the perianal skin, and pruritus.

The anorectal region is so heavily contaminated by a wide variety of organisms that the surgeon has always

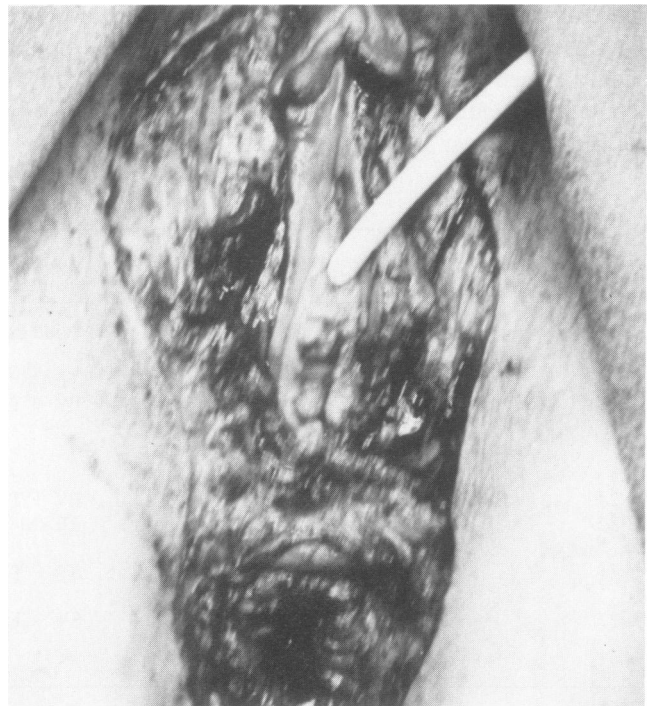


FIG. 4. Operative field showing extent of excision of vulvar and perianal skin. Foley catheter is in urethra above vaginal introitus.

been reluctant to cover wounds in this area. The surgeon's attitude toward wounds in the anus is one of conservatism. In 1953 Hughes⁶ successfully applied primary split-thickness skin grafts to anal wounds after laying open fistulae or excising fissures. In his experience the graft will take on a fresh wound no matter how contaminated, provided the two surfaces can be maintained in close apposition for several days. Infection will not develop unless a suitable medium exists in the form of necrotic tissue or hematoma.

Extending this philosophy to grafting the entire anal canal raised the theoretical question that anal continence might be lost or seriously compromised if the entire anoderm and its sensitive proprioceptive receptors were sacrificed. According to Goligher,⁵ anal continence depends on the acquired response to sensory stimuli of a complex of forces at the anorectal junction and in the anal canal. Skin of the perianal region and the modified skin below the dentate line exhibit sensibility to simple touch, pain, and heat and cold. There is a fairly widespread impression that the mucosa above the dentate line, like the lining of the rectum proper, is completely insensitive to ordinary tactile and painful stimuli.

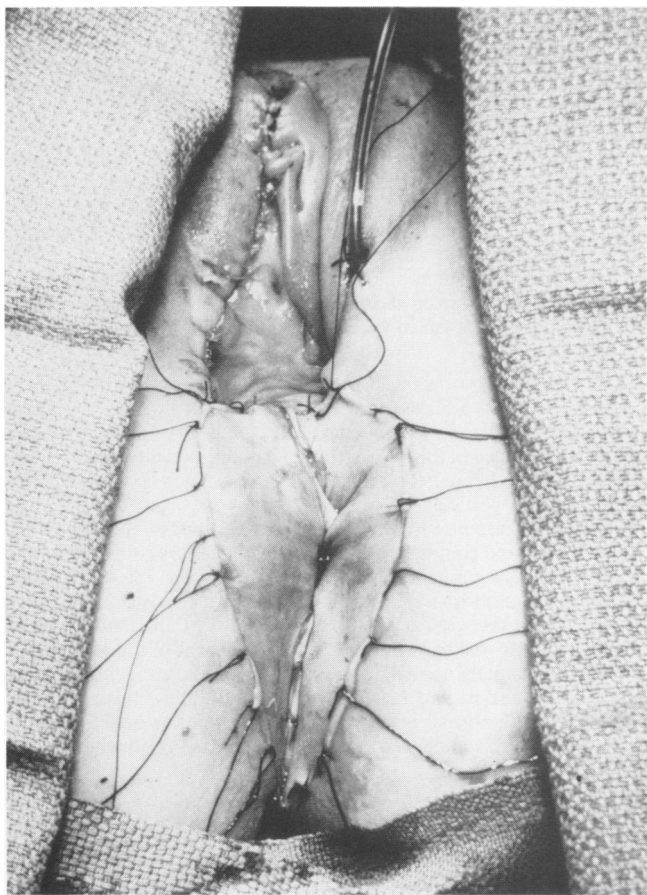


FIG. 5. Appearance of the skin grafts sutured in place prior to inserting the finger cot into the anal canal.



FIG. 6. Appearance of the anus and vulva 6 months after grafting.

In experimental studies by Goligher and Hughes,⁷ balloon distension of the rectum causes a feeling of fullness interpreted by the patient as a desire to pass flatus. The sensitivity of the rectum to this latter form of stimulation seems to be greater in the ampulla immediately above the anal sphincters than in the upper rectum. The rectum has the ability to distinguish between flatus and feces by differences in pressure. Flatus normally produces a lower degree of intrarectal tension than feces and the rectum is able to appreciate these minor differences of pressure. Although it may well be that this distinction depends, at least in part, on recognition by the rectum and upper

anal canal of other physical or chemical stimuli in its contents, in this study continence has been maintained in the absence of the highly specialized anal mucosa.

The anogenital region, comprising the cervix, vagina, vulva, perineum, and anus, has the potential for development of multicentric and multiple primary malignancies.⁸ Awareness of the existence of these tumors as a regional disease and the possible long intervals between successive malignancies should be born in mind. The presence of vulvar *in situ* carcinoma should arouse suspicion for a synchronous primary neoplasm elsewhere in this region. The frequent history of infection by the genital wart virus giving rise to condyloma acuminatum in the patients in this study gives plausibility to the theory that some of these viruses may act as a carcinogenic agent on this common epithelium.

Conclusions

Total excision of the anal mucosa with immediate split-thickness skin grafting is a highly satisfactory method of

treating *in situ* squamous carcinoma. This procedure has preserved anal function in all patients. At the present time, we would caution against the use of this procedure in patients with an invasive carcinoma until more experience has been gained.

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DISCUSSION

DR. H. WILLIAM SCOTT, JR. (Nashville, Tennessee): I want to congratulate Dr. Reynolds and his colleagues on developing a multidisciplinary approach that seems to be a very satisfactory solution to a problem that has been treated unsuccessfully very often in the past with radiotherapy and many varied and more radical operative procedures, including removal of the rectum, various and sundry modifications of the Whitehead procedure, plus removal of the perianal skin, and so on, that have resulted in an awful lot of grief for the patient.

I would like to point out also, though, that men can develop Bowen's disease of the perineum, and this is usually associated with perianal condylomata acuminata. Such a patient is now under treatment in the VA hospital in Nashville. At the time of his study and the biopsies of the squamous epithelium of the anal canal, the anus was both grossly and microscopically normal, and did not show any involvement, so a wide perianal excision of skin was carried out with grafting, similar to what Dr. Reynolds has described in women, and this patient temporarily has done fairly well.

But somewhat belatedly, on reexamination of his scrotum, he proved to have very tiny nodules that on biopsy proved to be *in situ* squamous carcinoma. He refused to have his scrotum skinned, so that he is now being treated with topical 5-fluorouracil. I wondered if Dr. Reynolds would comment on whether this is ever a satisfactory method of treating Bowen's disease, or whether this man is going to have to have scrotal skinning before he is cured.

DR. MAURICE J. JURKIEWICZ (Atlanta, Georgia): My own and the combined experience in our service with skin grafting the anus and perineum is limited to patients with (1) necrotizing symbiotic infections, (2) hidradenitis, and (3) burns. In these patients the grafts are invariably meshed, and the open technique is mandatory more often than not. In

most of the patients the grafting may go up to the sphincter, but seldom beyond, unless a portion of the anus itself is sloughed out.

To my knowledge, this is a unique series, in that the grafting extended to the dentate line and above. It is remarkable, not only that all the patients had a good take and that they regained continence to gas, fluid, and solids, but also that stricture or stenosis did not occur.

Two points, then two questions.

Wounds covered by skin grafts contract up to 60% of surface area. A mucosa-lined conduit of any sort, brought out to a skin interface, has a known high incidence of stenosis unless preventive measures are taken; indeed, often enough in spite of preventive measures: for example, the ileostomy stoma or the perineal pull-through.

Two questions: Is there something unique about the anal canal that prevents a skin graft from shrinking? What is your back-up procedure, should the skin graft fail to take, or partly take, and stenosis occur?

DR. JAMES J. MADDEN (Closing discussion): In answer to Dr. Scott's question, certainly 5-fluorouracil has been used in treating carcinoma *in situ*. In the vulva and in the perianal areas, it is very difficult to obtain patient compliance in this. This is a very painful modality, and in using this in the rectal canal, we felt that this would not be a satisfactory solution in this particular area.

I think—in response to Dr. Jurkiewicz's questions—that the entire problem is wound contraction. We have created a surgical defect in this situation that is a contractile, active muscle sphincter. We have chosen to reconstruct this with a split-thickness skin graft, and as Dr. Jurkiewicz points out, the skin graft certainly does contract. I think that the factor here is, basically, the daily normal bowel movement of the patient that allows us to maintain normal dilatation. We have chosen in this situation not to divert the patient before surgery, but to allow this patient to quickly regain a normal bowel habit, which I think is very important for the proper dilatation and maintenance of anal continence in this situation.