

Surgical Treatment of the Rectal Stage of Lymphogranuloma Venereum¹

ABDOMINOPERINEAL TRANSANAL RESECTION WITH PERINEAL COLOSTOMY AND PRESERVATION OF THE ANAL SPHINCTER
REPORT OF CASES

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THE MOST DREADED SEQUELA of infection with the virus of lymphogranuloma venereum is rectal involvement and its subsequent complications. Stricture of the rectum stands out as the most serious. The condition is more common in females, and this may be explained by the fact that the lymphatic drainage of the vulva and distal third of the vagina is directly into the perirectal and pelvic groups of lymph nodes, whereas that of the penis is into the inguinal lymph nodes, both deep and superficial. It is quite uncommon to see inguinal adenopathy in lymphogranuloma venereum of the female and relatively rare to observe pelvic and perirectal adenopathy with rectal lesions when the male is suffering from this disease. In all probability, the latter process in the male is the result of a direct initial lesion of the anorectal mucosa, be it incident to abnormal sexual relations (sodomy) or the result of an accidental implantation of the virus of the disease by means of infected rectal tubes or cannulae. Direct initial rectal involvement may occur in females also, but in most cases it follows the initial genital lesions, many of which pass unrecognized.

Kornblith² mentions the case of an eight-year-old white Puerto Rican girl, whose father had enlarged, suppurating inguinal nodes and whose mother had had a colostomy for rectal stricture. Both the girl and her parents reacted positively to the Frei test. The girl presented a primary rectal lesion, attributed to the use on her of the same enema tip utilized by her parents. Thus the initial lesion leading to a stricture may be rectal or extrarectal, in both cases the pathogenetic mechanisms being similar although the routes of establishment are different. In both cases the resulting pathologic changes are the same.

1. Received for publication July 20, 1943.

2. B. A. Kornblith, "Observations on Lymphogranuloma Venereum. Clinico-Pathologic Study of 60 Cases with Observations on the Histopathology of the Frei Test," *Surg., Gynec., & Obs.*, 63:99, 1936.

When a genital primary lesion has drained into the pelvic and perirectal lymph nodes, these become the seat of an inflammatory reaction similar in all respects to the one found in the inguinal nodes when they become involved. The pelvic and extrarectal networks of lymphatics become in turn involved; as the process advances into chronicity the rectal or intramural network of lymphatics also becomes diseased. The inflammatory reaction is characterized by proliferation in the affected tissues of fibroblasts and histiocytes. This fibroblastic proliferation gives rise to fibrosis with much induration, thickening, and contraction of the various coats of the rectal wall. The bowel becomes narrowed, and a stricture forms. Concomitantly, the mucosa becomes eroded at various places and finally, definite ulcerations develop. These are variously sized, ragged, irregular, rather deep and easily bleeding. As a rule, pyogenic infection becomes superimposed on the ulcers.

In our experience, the more advanced strictures, especially those of the tubular variety, are characterized by extensive ulceration of the mucosa throughout the extent of the strictured area but sharply demarcated from the grossly normal mucosa of the bowel proximally. We have observed that the mucosa distal to the stricture is granular, friable, edematous, hyperemic, and bleeds easily, and that it contains multiple small ulcers. We have also seen cases of rectoperineal fistulae with foul purulent fecal discharge; one of our cases had, previous to admission, a rectovaginal fistula that had healed spontaneously. Anal papillomatous growths have been frequently noted. All these manifestations, however, have been well described in the literature.

When the initial lesion takes place in the rectal mucosa, there is formed an ulceration on which pyogenic organisms thrive and multiply. The intramural lymphatics in the vicinity of the ulceration become involved, and the extrarectal and pelvic lymphatic chains also develop chronic grades of inflammation with fibrous tissue formation. The resulting pathologic changes are identical to the ones already described when the initial lesion is extrarectal.

According to Barber and Murphy,³ "fan-shaped masses of fibrotic tissue interspersed with necrotic foci, evidently disintegrated lymph nodes, were observed to point from the lateral walls of the mid-rectum beneath the deep fascia and close to the periosteal surfaces of the floor of the true pelvis." In their cases they noticed varying

3. W. H. Barber and W. Murphy, "Lymphogranuloma Venereum," *Ann. Surg.*, 113:30, 1941

degrees of lymphadenitis within the abdomen, from "barely palpable nodes in the presacral area to general enlargement of the retrocolic nodes of the sigmoid and descending colon." We observed only slight pelvic and pericolonc adenitis.

According to their extent, strictures have been described as annular or diaphragmatic and cylindrical or tubular. Kornblith⁴ states that 75 percent are tubular and the rest diaphragmatic; he has found the site of stricture to be limited to the lowest 8 cm. of the rectum. Our series shows four diaphragmatic and five tubular strictures. The lesions start anywhere from 3 to 6 cm. from the anal opening and extend for an average distance of 7.5 cm., the shortest tubular one being 4 cm. and the longest, 11 cm.

Marino⁵ describes a case where the entire rectum, sigmoid, and descending colon were stenosed, with infiltration of all the coats. Palmer, Kirsner, and Rodanische⁶ mention another in which there were two strictures, one in the rectum and one in the mid-portion of the sigmoid. They also point out the interesting fact that in four of their patients with stricture "the rectal mucosa was friable, edematous, and indistinguishable from that of patients with non-specific ulcerative colitis." We have a similar case under observation. Llombart and Mañero⁷ write of a case with a positive Frei test, rectal stricture, and multiple strictures of the ileum, which they attribute to lymphogranuloma venereum. We also recall a case, operated on about two years ago, in which two closely adjacent granulomatous, ulcerated, bleeding lesions were encountered in the proximal jejunum. They were successfully resected (approximately 12 cm. of jejunum were excised followed by end-to-end anastomosis). The patient gave a strongly positive reaction to the Frei test and the histologic picture of the lesions was compatible with a diagnosis of lymphogranuloma venereum.

The most characteristic microscopic pathologic lesion is believed to be the small focal abscess with central necrosis and peripheral epithelioid and giant cells of the Langhans type. Our material did not show these abscesses, but frequent epithelioid and occasional

4. B. A. Kornblith, "Lymphogranuloma Venereum—Surgical Aspects," *Am.J.Dig.Dis.*, 6:712, 1939.

5. A. W. M. Marino, "The Anorectal Phase of Lymphogranuloma Inguinale," *Ann.Surg.*, 102:1086, 1935.

6. W. L. Palmer, J. B. Kirsner, and E. C. Rodanische, "Studies on Lymphogranuloma Venereum Infection of the Rectum," *J.A.M.A.*, 117:517, Feb. 14, 1942.

7. A. Llombart and J. Mañero, "Sur la localisation de la maladie de Nicolas-Favre sur l'intestin grêle. Stenosis ileales multiples," *Ann.d'anat.path.*, 16:597, 1939.

giant cells were observed. The other changes in our material, described by the pathologist Dr. Enrique Koppisch, are those of a chronic granulomatous process, namely, focal or perivascular lymphocytic and plasma cell infiltration throughout the coats of the bowel, especially the submucosa, edema, abundant fibrosis and scarring, and replacement of the mucosa by granulation tissue.

REVIEW OF THE TREATMENT GIVEN IN RECTAL STRICTURE

A review of the literature as far back as 1930 reveals a general agreement with the fact that the advanced pathologic changes, accompanying the anorectal syndrome, respond to no other treatment but surgical. Palmer *et al*⁸ report a case in which an early stricture disappeared almost completely after treatment with four daily grams of sulfanilamide for ten days. However, since there was no adequate follow up of this patient, the permanency of such a cure remains questionable. We have tried this therapy without being able to influence the stricture itself.

Surgical procedures tried in rectal strictures can be classified as follows:

(a) *Mechanical dilatation*. This is performed with or without anaesthesia. It is the simplest of the surgical methods and represents one of the oldest attempts to deal with the strictures. Dilatations are contraindicated in acute proctitis and proctitis because of the danger of causing an exacerbation and marked systemic manifestations. It has been suggested that they may be of help in the less fixed diaphragmatic strictures. Edwards⁹ believes that many patients can be carried along for years with little inconvenience by periodic dilatations. However, our experience shows that, with very few exceptions, they either become gradually worse under such treatment or else tire of the procedure by failing to follow treatment or if doing so, follow it only irregularly.

The exception that proves the rule—Case U. H. A-2500—is worth mentioning here. The patient was a 30-year-old woman whose case has been followed up for five years. On admission to the Out-patient Department, she complained of intestinal trouble, loss of weight and strength for seven years. She then weighed 86 pounds and was running a slight temperature. The Frei test proved definitely positive; rectal examination showed the typical stricture

8. W. L. Palmer *et al*, *op. cit.*

9. M. Edwards, "The Management of the Ano-Rectal Syndrome of Lymphogranuloma Inguinale," *South.Med.J.*, 30:1194, 1937.

with changes of the mucosa as previously described. In spite of gradual rectal dilatations and Fuadin (antimony), intramuscularly, the patient continued to lose weight until she had reached 73 pounds. She also showed definite signs of a psychoneurotic nature, with frequent hysterical outbreaks.

Neoprontosil (gr. X t.i.d.) was prescribed and gradual rectal dilatations were continued. Despite a Bartholinitis, which had appeared on and off and which had finally drained spontaneously and ceased altogether with sulfa therapy, her general condition improved. Eighteen months after admission to the Out-patient Department she weighed 93 pounds and was moving her bowels satisfactorily with the aid of mineral oil. Prontosil was necessarily interrupted and hematinics and tonics were given instead. Rectal suppuration and bleeding disappeared; her mental condition also improved. Three diathermy treatments were added but the patient would not come regularly to her appointments and these had to be discontinued. In 1941 sulfathiazole was substituted for Neo-Prontosil. The patient took it on and off, though never with regularity. Rectal dilatations were continued at very irregular intervals. On her last visit to the clinic, October 20, 1943, she weighed 118 pounds; she looked healthy and her bowel movements were regular when she took the prescribed mineral oil.

Despite such remarkable improvement, on every visit to the clinic this patient begged for an operation which we always refused, as we did not think that surgery could accomplish any more than conservative measures had. Furthermore, we believed that she was the type of patient who would not tolerate radical surgery well.

(b) *Proctotomies*. These may be internal, radial, or posterior. The earliest proctotomy for rectal stricture on record was that of Dupuytren who performed four radial incisions with a bistoury in 1854 and followed them by dilatations. Lockhart-Mummery and Lloyd¹⁰ have said: "Internal proctotomy and subsequent dilatations are excellent and permanent if the patient will endure the inconvenience of dilatations long enough to counteract the contraction of scar tissues." We believe that proctotomies are only temporizing procedures and are to be condemned because of the scarring they produce.

(c) *Incision or excision of fistulous tracts, condylomata, polyps, and papillomas*. These procedures are more or less futile, except that an

10. J. P. Lockhart-Mummery and D. O. V. Lloyd, "The Operative Treatment of Fibrous Strictures of the Rectum," *Brit. J. Surg.*, 23:19, 1935.

occasional case without much stricture might benefit greatly. See case R-6.

In January, 1938, we (J.N.B.) first saw this patient at our private office. He had been suffering from severe constipation and bloody and purulent discharge per rectum for five years. He had also lost 25 pounds and his weight was then down to 115. There were two papillomas and one small fistulous tract at the anal border, a rectitis, and a beginning stricture. The patient was running quite a high daily temperature, which had gone up to 37.5° C, or 38° C, for several years; lately the daily rise had been as high as 39.5° C. He could hardly keep up with his work. The patient was hospitalized, given Neo-Prontosil which reduced his temperature, and the fistula was incised and drained; the papillomas were excised. He remained afebrile for about one week and then the fever and the malaise reappeared despite satisfactory local conditions. Neo-Prontosil therapy was resumed and the fever disappeared. His convalescence was uneventful after that.

For three years thereafter the patient was given neoprontosil and prontosil on and off, aided by frequent rectal dilatations. A small fistula formed during a relapse but, as he was going to New York City at the time, he was referred to Dr. A. V. Burt, under whose guidance we had had our proctological training, years before, at the Vanderbilt Clinic. Dr. Burt incised and drained the fistula under local anaesthesia, advising him to continue Neo-Prontosil therapy with frequent hematological check-ups. The patient gained strength again; his weight went up to 150 pounds; he looked the picture of health. A slight incontinence remained due to a defect in the sphincter whenever his bowels were loose, but the stricture apparently had disappeared. He occasionally resorted to sulfathiazole for a flare-up of the proctitis but this condition has appeared less and less frequently of late. The patient has kept up his weight, his health, and his work.

Surgery of fistulous tracts leaves chronic sinuses, and other lesions recur simply because the underlying pathologic process is not attacked. Before coming to us, one of our cases had had two unsuccessful plastic operations for a recto-vaginal fistula which, strikingly enough, healed spontaneously in course of time.

(d) *Evacuation of the perirectal space and retroproctitic mobilization of the stricture*. This technique has been advocated by Gómez.¹¹ We

11. D. Gómez, "Nueva orientación en el tratamiento de la rectitis estenosante," *Arch. de Med., Cir., y Espec.*, 37:1005, 1934.

consider it ineffectual and potentially dangerous inasmuch as it might stir the infecting virus which is latent in the endopelvic fascia and perirectal tissues, thus exacerbating the condition.

(e) *Temporary colostomy*. This procedure is seldom used and then only as an emergency operation in cases of acute colonic obstruction due to a lymphogranulomatous stricture. Supportive measures, decompression with the Wangenstein suction apparatus, enemata, and careful rectal dilatations should be tried first.

We do not favor such a procedure for the temporary isolation of the distal segment. Advocates of this method claim that the rest thus afforded the distal segment (which can then be irrigated thoroughly and gradually dilated) permits healing of the lesions, after which the fecal stream can be re-established. We seriously question the so-called healing of the distal segment, even if sulfonamides are administered. We believe that improvement, if any, will be only temporary and that the condition will again become exacerbated as soon as a normal bowel continuity is re-established.

(f) *Permanent colostomy*. The first colostomy recorded in medical literature was performed by Littré in 1710. Permanent colostomy has been frequently performed as a palliative measure in the treatment of lymphogranuloma venereum even when the true nature of the stricture was not recognized. Marino,¹² Kornblith,¹³ Barber,¹⁴ Borjas,¹⁵ and Edwards¹⁶ have described the results to be expected in certain cases by the use of this procedure.

Borjas generally advocates the colostomy at the descending colon, followed by careful postoperative treatment with sulfonamides and irrigation of the distal segment. He says: "Selection of the site of colostomy depends on x-ray studies and on ample exploration of the colon at celiotomy." If there is no local improvement, he often resorts to perineal resection. Borjas is not in favor of the Mickulicz double-barreled type of colostomy because of prolapse of the barrels and the lack of control of bowel function. He employs the Kurzhahn-Cuneo colostomy as modified by Corachan and Baquero,¹⁷ in which the proximal cut end of the bowel is brought out through a skin

12. A. W. M. Marino, "The Anorectal Phase of Venereal Lymphogranuloma," *Am.J.Surg.*, 45:293, 1939; A. W. M. Marino, *op. cit.* (5).

13. B. A. Kornblith, *op. cit.* (4).

14. W. H. Barber and W. Murphy, *op. cit.*

15. A. Borjas, "La enfermedad de Nicolas-Favre o cuarta enfermedad venérea. Contribución al estudio del tratamiento quirúrgico de sus localizaciones ano-rectales." *J.Internat. Col.Surg.*, 5:50, 1942.

16. M. Edwards, *op. cit.*

17. Corachán and Baquero, cited by A. Borjas, *op. cit.*

tunnel and clamped with a special metal device. Most surgeons, however, prefer the double-barreled colostomy.

Edwards¹⁸ is also in favor of determining the site of colostomy by exploratory laparotomy. Jaffé,¹⁹ of the University of Venezuela, reported seven out of thirty-seven autopsy cases in which the lesions were found as high as the sigmoid. Pérez Carreño²⁰ observed three cases of invasion of the sigmoidal colostomy by upward extension of the lymphogranulomatous process.

In our opinion, colostomy does not solve the problem of lymphogranulomatous rectal stricture. Aside from the physical and psychological invalidism it creates in the patients (most of whom object seriously to the procedure), the site of colostomy has the disadvantage of being liable to involvement by direct extension of the disease proximally along the bowel. Furthermore, the general condition of the patient does not always improve after colostomy, for the diseased focus in the lower bowel segment remains more or less active and toxic products continue to be absorbed from this area, thus perpetuating the state of general ill health. Our experience shows that rectal suppuration and an irritating discharge frequently persist in spite of the colostomy.

(g) *Perineal resection with sacral anus*. This procedure is advocated by Borjas²¹ when the colostomy fails to produce definite improvement. The Kraske type of operation is then performed. In 1933 Yeomans²² reports a series of perineal resections for lymphogranuloma venereum; Barber and Murphy²³ also report a few cases thus treated, and point out the rapid healing of the wound in such cases. We believe that by a simple perineal approach even the most experienced surgeons will find tremendous technical difficulties in the liberation of the diseased segment of bowel.

(h) *Whitehead operation*. Hartman²⁴ reports thirty-four cases of rectal stricture in 1922, operated by the Whitehead technique. Only two deaths occurred in the series with the remaining thirty-two cases being classified as successful. There was no adequate follow up, however. Considering the extensive nature of the pathologic process in the rectal lesions of lymphogranuloma venereum, it is obvious

18. M. Edwards, *op. cit.*

19. Jaffé, cited by A. Borjas, *op. cit.*

20. Pérez Carreño, cited by A. Borjas, *op. cit.*

21. A. Borjas, *op. cit.*

22. Yeomans, cited by M. Edwards, *op. cit.*

23. W. H. Barber and W. Murphy, *op. cit.*

24. Hartman, cited by M. Edwards, *op. cit.*

that the limitations of the Whitehead procedure make it totally inadequate.

(i) *Abdominal colostomy with perineal excision.* Miles was among the first to use this procedure for carcinoma of the rectum, when a single-barreled left inguinal colostomy and a perineal excision were performed in one stage. Later on, Lockhart-Mummery employed this same procedure in one and two stages for malignancy of the rectum or sigmoid, but favored internal proctotomy and dilatations for venereal rectal strictures.

Bacon²⁵ and Bacon and Griffin²⁶ also used the abdominal colostomy and perineal excision in a one-stage operation for certain cases, but prefer the two-stage procedure, irrigating the lower loop daily with warm potassium permanganate for three to six months between the first and second stages. Bacon reports twenty-four cases thus treated without any mortality. Convalescence, however, was protracted and invalidism fairly marked, following proctectomy. In seven cases subsequent contracture of the skin occurred around the colostomy. "An average period of hospitalization following the first stage was twelve days and, following the second stage, twenty-nine days." We are opposed to the abdominal type of colostomy and consider that the one-stage procedure, if properly carried out, is quite safe and time-saving.

(j) *Abdominoperineal resection with abdominal colostomy in one or two stages.* At the turn of the century, Miles was among the pioneers who developed the technique for this operation, which was intended primarily for carcinoma of the rectum and later employed for lymphogranulomatous rectal strictures. Barber and Murphy²⁷ mention eighteen cases in which they performed abdominosacroperineal operations; four of them died postoperatively, giving a mortality rate of about 22 percent. However, those patients followed up gained in weight, felt very much improved, and learned to control the abdominal colostomy. Morris²⁸ reports the case of a 43-year-old white male on whom abdominoperineal resection had been performed "because of the desire to remove all active foci." The first stage consisted of plastic repair of a previously existing colostomy

25. H. E. Bacon, "The Surgical Treatment of Lymphogranulomatous Strictures of the Rectum. Report of 24 Cases," *South Med. J.*, 34:31, 1941.

26. H. E. Bacon and O. P. Griffin, "Lymphogranuloma Venereum with Special Reference to Rectal Stricture," *Am. J. Surg.*, 56:166, 1942.

27. W. H. Barber and W. Murphy, *op. cit.*

28. J. H. Morris, "Abdominoperineal Resection of the Rectum for Lymphogranuloma—Case Report," *Ann. Surg.*, 3:152, 1940.

that was strictured. In the second stage the sigmoid and rectum were mobilized from above and the operation completed by perineal removal. This procedure is well known in Venezuela, where the Corachán-Baquero colostomy is followed in the same stage, or in a second stage, by abdominoperineal resection.

Edwards²⁹ prefers perineal, or abdominoperineal, resections with abdominal colostomy to abdominoperineal procedures in which the perineal resection is performed transanally, and the sphincter preserved. Though he does not seem to have had first-hand experience with this method, he is afraid that such a procedure may fail to remove adequately all the involved tissue.

The main objection to the abdominoperineal procedure, with abdominal colostomy, is the type of anus established. This, in our opinion, is difficult to control and is found highly objectionable by the patients. Furthermore, we believe that intraabdominal division of the gut, even when all precautions are taken, entails greater danger of contamination than the method favored by us wherein the bowel is divided outside the body after the diseased portion has been delivered through the perineal wound.

(k) *Abdominoperineal resection (proctosigmoidectomy) in one stage with perineal anus.* This technique was fully described by Wayne Babcock in 1932, and since then he has been an ardent advocate of the method for carcinoma of the rectum or rectosigmoid. The operative details are well explained in his original article, hence it is only necessary to say now that (1) he saves time by omitting peritonealization of the pelvic floor; (2) he splits the sphincter ani, preferably posteriorly; (3) he brings the cut end of the bowel out through the sphincter; (4) he does not suture the bowel to the latter, and (5) he leaves a rectal tube in place for a few days. Babcock has apparently not used this procedure in cases of lymphogranulomatous rectal stricture or, at least, he has not made a report thereon.

In France, Villard and Ricard³⁰ were the first to report on the use of the abdominoperineal resections with perineal anus for venereal rectal stricture. Dmitri and Grigorescu³¹ followed in 1939 with a report of nineteen cases thus treated, wherein they performed the perineal part of the operation transanally and conserved the sphincter, as advocated by Babcock. They also peritonealized the raw areas and formed a new pelvic floor. There was one death in this

29. M. Edwards, *op. cit.*

30. Villard and Ricard, cited by M. Edwards, *op. cit.*

31. Dmitri and Grigorescu, cited by M. Edwards, *op. cit.*

series, while seventeen were reported cured and one improved. However, we cannot say whether or not there was an adequate follow up.

Since the latter part of 1941, Dávila³² has been employing a procedure that is an improvement on the Babcock technique. He calls it a "simultaneous one-stage abdominoperineal resection with conservation of the sphincter." At the onset, the patient is placed in the lithotomy position and prepared for both the abdominal and perineal parts of the operation, which are then performed simultaneously by two teams of operators. The perineal procedure is started as soon as the bowel is found mobilizable at laparotomy. Dávila usually peritonealizes the pelvic floor. The perineal dissection is performed through an elliptical incision, enclosing the anus and extending posterior to it, after the anal opening has been purse-stringed. Care is taken to leave intact the anterior attachment of the sphincter ani, which muscle is well dissected out from the perianal tissue. The bowel is brought down after both the division of the levatores ani and the perirectal dissection have been accomplished and the rectal connection to the sphincter severed. The bowel is then brought out through the sphincter, clamped just proximal to the tape ligature and just distal to the sphincter, and the distal segment removed with the cautery twenty-four hours later. Dávila has already performed over thirty such operations for rectal stricture.

PROPOSED OPERATION

We consider the Babcock operation, as modified by one of us (J.N.B.), ideally suited for the radical surgical treatment of rectal stricture due to the virus of lymphogranuloma venereum. In this connection we have chosen the continuous, fractional or intermittent, method of spinal anaesthesia, as first described by William T. Lemmon. This method has always given us very satisfactory results, since it is gratifying to be able to carry out this long, involved operation without the employment of a general anaesthetic.

After the patient is in the lithotomy position with the flexible spinal needle *in situ*, both abdominal and perineal operative fields are thoroughly washed with tincture of green soap and benzene, and then painted with tincture of mercuric chloride. Sterile leggings and sheets are used to drape the patient so that the lower abdomen

32. B. Dávila. Personal communication.

remains exposed and the perineal region temporarily covered by sterile sheets. The abdominal part of the operation is then started by a midline incision extending from the umbilicus to the symphysis pubis, through skin, subcutaneous tissues, and linea alba. The recti are retracted laterally and the peritoneum incised from above downwards. Care is taken not to injure the bladder which has been emptied by catheterization just before the patient is brought to the operating room. The excellent abdominal relaxation produced by the spinal anaesthesia, together with the midline approach, afford a very adequate exposure of the terminal descending colon, sigmoid, and rectum.

The operative area is thoroughly isolated with warm towels and the sigmoid displaced medially, fan-like, so as to be able to incise the lateral leaf of the mesosigmoid. Blunt dissection with sponges on ovum forceps and sharp dissection with scissors is then carried downwards along the divided leaf as far as the brim of the pelvis. A similar procedure is performed along the medial leaf of the mesosigmoid, after the sigmoid has been displaced laterally in fan-like fashion. Great care is also taken not to injure the left ureter and the left ovarian or spermatic artery exposed during the course of this dissection. The inferior mesenteric vessels are identified as they course in the mesosigmoid, ligated with No. 30 cotton thread, divided between clamps, and again similarly ligated. Blunt and sharp dissection is also carried down to the pelvic brim on the lateral aspect of the mesosigmoid. The sigmoid will then be found quite free and movable. (In all our cases mobilization of the rectosigmoid was possible in spite of dense perirectal adhesions and granulation tissues in some, which made the procedure laborious.) Traction is made on the sigmoid and blunt and sharp dissection extended along the hollow of the sacrum so as to free the rectum posteriorly as far down as the tip of the coccyx. The dissection is then undertaken on both sides of the rectum, eventually dividing the lateral rectal ligaments.

In spite of previous ligation of the inferior mesenteric vessels, bleeding is at times profuse because of adhesions and granulation tissue. Hot towels and sponges are used liberally to control this bleeding. Finally, attempts are made to liberate the rectum anteriorly as far down as possible: dissection is undertaken between the vagina and uterus and rectum, or between the prostate and seminal vesicles and the rectum. Dissection is more difficult and dangerous in this region, mobilization being usually completed with

more safety through the perineum. During this last process one tries to include as much perirectal tissue as possible: first, because as much diseased tissue as possible should be removed and second, in order to avoid injury to the bowel wall. The sigmoid is again inspected and a level selected proximal to the uppermost extension of evident gross colonic involvement. If this level can be pulled with ease along the sacrum and if it reaches the lower end of that bone without tension, we feel that resection is safe at that point. A strong sterile tape is then securely tied around the gut just below the level selected for resection and the tape tucked as far down as possible into the hollow of the sacrum. The sigmoid is then similarly placed in that area. No peritonealization of raw areas and no creation of a new pelvic floor are performed since we consider them unnecessary and time-consuming.

The sterile towels are now removed and the operative field examined for assurance that there is no bleeding. The abdominal wall is closed "en masse" with interrupted through and through mattress No. 30 cotton thread sutures, and the skin borders are brought together further by intercalating single interrupted sutures of No. 60 cotton thread. No drainage is established. Dressings are applied and the perineum uncovered in order to proceed with the second stage of the operation.

The anus is then closed with a double No. 30 cotton thread purse-string suture and the threads are left long for traction purposes. A circumanal skin incision is made just inside the sphincter ani and dissection with scalpel and scissors carried cephalad along the plane thus created. The sphincter, or what remains of it, is carefully preserved and gradually dilated and stretched so that it continues to lie peripheral to the plane of dissection. The levatores and other perineal muscles are divided; the dissection is deepened until the hollow of the sacrum is reached. The sterile tape is identified by palpation and gently but firmly pulled out until the tied bowel is delivered through the dilated sphincter. Traction is then made on the rectum and the remaining anterior rectal adhesions are sharply divided, taking care not to injure the posterior vaginal wall in the female or the urethra and prostate in the male. As the rectum is freed, the anus, rectum, and distal sigmoid are brought to the outside. Again light traction is made on the exposed sector of bowel so that the proposed level of resection at the sigmoid lies just outside the preserved sphincter fibers. That level is then sectioned with the cautery between small Payr clamps and the seromuscular layers

of the cut end of the sigmoid united without tension to the sphincter ani by interrupted No. 60 cotton thread single sutures, radially disposed. The Payr clamp on the cut end of the sigmoid is then released, whereupon the bowel is usually seen to retract until it lies flush with the anus. The perineal dead space posterior to the newly created anus is then packed with iodoform gauze and a sterile pad applied against the anus and held in place by a "T" binder.

In those cases where the bowel has been accidentally injured during the course of the dissection, six to eight grams of sulfathiazole powder are insufflated, half into the pelvic cavity before closing the abdomen and half into the perineal dead space before the packing is inserted. At the end of six to seven days, the perineal packing is removed unless edema of the perineum supervenes, when it is removed earlier. Abdominal sutures are removed from the eighth to the tenth day.

This technique offers the following advantageous features: (1) use of the continuous spinal method of anaesthesia; (2) convenience afforded by having patient ready in the lithotomy position from the onset of the operation, thus avoiding loss of time between the abdominal and perineal stages; (3) avoidance of peritonealization of raw areas or formation of a new pelvic floor; (4) closure of the abdominal wall with through and through sutures; (5) use of cotton thread as a cheap and dependable suture material for this type of surgery; (6) transanal approach with preservation of the sphincter, thus establishing a much more physiological artificial anus than that offered by abdominal colostomy or by perineal procedures that would divide it, and (7) radical removal of as much diseased tissue as is possible and is compatible with the physiological function and rehabilitation of the patient.

DISCUSSION OF CASES

So far, nine cases have been treated by a one-stage abdomino-perineal resection with conservation of the sphincter ani.

The youngest patient was 25 years and the oldest 48, with an average of 37.7 years for the whole series. Eight were females and one was a male; of the women five were colored, three were white. The male was also white, single, and a sodomist. Three of the females were married, two were separated from their husbands, two were widowed, and one was a concubine. Rectal symptoms had been complained of from one year nine months to eleven years, with an average of 4.6 years.

showed anal papillomata. The sphincter tone was considered fairly good in one case, the surgical specimen of which did not show true stricture on pathologic examination. It was fair in three, poor in two, and totally lacking in the remaining three cases. Hemorrhoids were observed in two patients. Rectal examination was severely to moderately painful in all. The rectal mucosa showed extensive ulcers that bled easily in all cases except one: the patient whose stricture was not found pathologically did not have any. At examination all nine cases presented the typical "bag of beans" feeling described by other authors, that is, a granular feeling to the mucosa distal to the stricture. Ulcers were visualized at the site of stricture with the aid of a children's proctoscope. In three cases the mucosa appeared edematous. Thickening and fibrosis of the rectal wall gave an unyielding sensation on digital examination.

All strictures were pronounced—except in the case (A-1735) referred to above, which had only a moderate diminution in the caliber of the lumen—and were found to start anywhere from 3 to 6 cm. from the anal opening. Four (including the male) were classified as circular (diaphragmatic) and the remaining five as tubular. The former were those that did not extend for more than 3 cm. longitudinally, while the latter varied in extension from 11 cm. to 4 cm., with an average of 7.5 cm. These findings were corroborated on pathologic examination.

Laboratory findings

The outstanding laboratory determination in lymphogranuloma venereum cases should, of course, be the Frei test. All the cases in this series gave a positive reaction, ranging in intensity from 2+ to 4+. Tests were read from forty-eight to seventy-two hours after the injection of chick embryo antigen with control. We remember very distinctly one severe reaction consisting of a large papule measuring 2 cm. in diameter that developed pustulation, with an extensive crop of large vesicles and vesicopapules that first surrounded the lesion and then spread all over the forearm on an edematous, inflamed, reddened base. There was moderate pain and fever. This condition, however, cleared up in about a week's time.

The average red cell count was 3.91 million with values ranging from 2.8 to 5.02 million. The hemoglobin averaged 69.6 percent with a range of 53 to 91 percent. Total white cell counts fell within normal limits. The differential counts were interesting, since five cases on admission showed an absolute lymphocytosis which ranged

from 36 to 56 percent and averaged 49 percent in these five cases. A search of the literature has failed to show similar reports.

Urinalyses were essentially negative. The stools of one patient (A-200) showed the presence of *S. mansoni* ova on routine examination.

Serum protein determinations were carried out in six cases, total proteins ranging from 7.42 to 8.51 gm. percent with an average of about 8 gm. percent. The globulin fraction varied from 2.59 to 4.84 gm. percent with an average of 3.76. There was a reversal of the albumin-globulin ratio in two of the cases (globulin 4.70 gm., albumin 3.50 gm., and globulin 4.84 gm., albumin 2.58 gm.).

Of seventy-three cases which they studied, Jones and Rome³³ found that 64 percent had total proteins above 8 gm. percent and that 90 percent had globulin above 3 gm. percent. In 32.5 percent the albumin was less than 4 gm. percent. Fifty-two percent had reversed A:G ratio due to an increase in globulin, with or without a decrease in the albumin. The Takata-Ara test was positive in 40 percent of the cases. Hernández Morales³⁴ has found that many cases of lymphogranuloma venereum give a positive cephalin test.

Nonprotein nitrogen of the blood, determined in seven cases, showed values within normal limits. The sedimentation rate was determined in seven patients and was found moderately high in two females and more so in the male patient. Four females had a normal sedimentation rate.

Only one female patient showed a positive Kahn.

X-ray studies

Eight cases were examined radiologically after a barium enema when rectal strictures were demonstrable. The rectal ampoule was quite distorted in two cases. Even the case that did not show the stricture pathologically was considered positive roentgenologically. In this particular case (A-1735) spasticity of the colon was also noticed. Wien³⁵ reports a similar case in which x-ray revealed an apparent rectal stricture that was not found at autopsy.

Preoperative period

The average number of hospitalization days prior to operation

33. C. A. Jones and H. P. Rome, "Lymphogranuloma Venereum as a Systemic Disease," *Internat. Clin.*, 2:179, 1938.

34. F. Hernández Morales, Personal communication.

35. M. S. Wien, "Inguinal Lymphogranuloma and Its Relation to Stricture of the Rectum," *Arch. Path.*, 19:331, 1935.

was 13.8, the longest period being 24 and the shortest, 8. Sulfathiazole in moderate doses was administered preoperatively to two patients. A few days after the onset of therapy one of them, the male patient, developed a toxic erythema, nodular in type, accompanied by reddened, painful, swollen finger tips, and the drug was discontinued. The other patient tolerated the drug well. Another case was given neoprontosyl without any reaction. Three of the patients were on a low residue diet, five on a regular diet, and one on a soft diet. Transfusions were given preoperatively to three of the patients and hematinics (ferrous sulfate, ferric ammonium citrate and liver extract) to most of them. Laxatives, purgatives, and enemas were ordered as required.

While in the hospital, one of the females developed subacute partial intestinal obstruction, which was treated conservatively, the condition subsiding rapidly. Another patient, who had a history of joint swellings and pain, developed swelling and pain of the right knee. Aspirin and iodides were given orally and ice bags placed on the swollen joints. As said before, this condition was considered part of the picture of lymphogranuloma venereum.

Anaesthesia and operation

(a) *Anaesthesia.* Procaine hydrochloride crystals, dissolved in spinal fluid to make a 5 percent solution, was the anaesthetic used in all cases and was given by the so-called continuous, fractional or intermittent, method. The total amount used in each case varied from 175 to 350 mgm., with an average of 264. An initial dose of 100 mgm. was given in every case with successive doses of 50 mgm. each, as needed, every fifteen to forty minutes. Anaesthesia was excellent in all cases but one, when supplementary N₂O and ether anaesthesia were added. This was necessary because of a leakage of the spinal fluid around the indwelling needle due to malposition of the latter. There were no reactions attributable to the anaesthesia.

(b) *Medication.* Three cases were transfused during the operation; seven received infusions of 10 percent glucose-in-saline. Eight were given 50 mgm. of ephedrine hypodermically when the blood pressure showed a tendency to drop.

(c) *Operation (modified Babcock procedure).* This was performed according to the one-stage combined abdominoperineal resection with perineal anus and conservation of the sphincter ani, already described. In three cases there was gross contamination of the abdominal cavity due to accidental tears in the rectum. Sulfathiazole

zole powder (6 to 8 gm.) was insufflated into the pelvic cavity, perineum, and wound layer in five cases, including those where gross contamination had taken place.

The operations lasted from one hour and twenty-two minutes to three hours and thirty-five minutes, but the average time was two hours and six minutes. Slight to moderate shock was present in eight cases, sometimes during the operation. The male patient went into severe shock but recovered uneventfully.

Surgical pathology

This procedure confirmed the clinical findings with the exception of case A-1735, where no definite stricture of the rectum was found even though there was some narrowing of the lumen. Perirectal adhesions and fibrosis, edema, thickening and induration of the rectal wall were evident in all cases. Grossly, the mucosa appeared extremely ulcerated, such ulceration covering all the area of stricture. In all cases but one the anal sphincter was atrophied in varying degrees, even in the case that did not show stricture. The involvement of tissues by the lymphogranulomatous process extended above the peritoneal reflection in some cases, and hence the length of bowel resected varied considerably perforce. The longest specimen measured 28.5 cm. and the shortest, 14 cm.; the average length of the surgical specimens was 20.3 cm.

Both grossly and microscopically the degree of perirectal lymph node involvement observed was slight. This was striking, inasmuch as the pathogenesis of the condition would make one anticipate extensive changes in the nodes.

Granulation tissue formation and perivascular infiltration with plasma cells and lymphocytes were constant elements. Edema of the submucosa and fibrosis with scarring of the muscularis were also present. Typical necrotic foci, as described in the literature, were not found in this material, but a systematic search was not made.

Postoperative period

Hospitalization ranged from 37 to 13 days with an average of 28. The longest febrile period lasted 28 days (the case developing an abscess in the left lower quadrant) and the shortest, 2. The average febrile period was 13 days long; the highest temperature recorded, 104° F. (abscess case).

Wound healing

(a) *Abdominal wound.* All wounds healed by primary union with

exception of one case sutured "en masse," in which abdominal distention and forceful vomiting occurred secondary to a paralytic ileus. These complications brought about a partial disruption of the wound layers at about the middle of the incision, with hematoma formation. The disrupted area was resutured with through and through cotton thread sutures after evacuation of the hematoma and the wound healed successfully.

(b) *Perineal wound.* The dead spaces created by the perineal dissection granulated uneventfully in all cases. There was edema of the perineum in four. One case presented a slight sloughing of the posterior third of the perineum; in two there was some sloughing of the implanted end of the sigmoid which separated spontaneously in one case and needed trimming in the other.

Abdominal sutures and the perineal iodoform gauze drains were removed, on an average, between the sixth and the ninth days, but this time limit did not include the cases with perineal edema, where the drain was removed as soon as the edema revealed itself.

Complications

Two cases presented slight superficial wound infections due to the hemolytic *Staphylococcus aureus*. One of them had a small stitch abscess and the other several. One of the cases, in which gross contamination had occurred at the time of operation, developed a probable abscess in the left lower quadrant of the peritoneal cavity some two weeks later but it underwent resolution under sulfathiazole therapy.

None of the other patients developed peritonitis. Three of them had moderate degrees of paralytic ileus sometime during the first postoperative week but responded well to Wangensteen suction through the gastric tube and to supportive therapy.

One case developed a pyelonephritis during the second postoperative week, produced by a combined nonhemolytic streptococcus and a nonhemolytic staphylococcus. This condition was treated with sulfathiazole and, later, with urotropine. Another case, complicated by the presence of a caruncle and a stricture of the external urethral meatus, developed a severe cystitis during the second postoperative week. It, too, responded to urotropine and tincture of hyoscyamus together with a high fluid intake.

Three cases receiving sulfathiazole postoperatively developed toxic erythema of the nodular type and painful, reddened, swollen

finger tips. A fourth presented the latter but no erythema. All cases recovered spontaneously after the drug was discontinued.

An interesting finding appeared in two of the cases during the course of the sulfathiazole therapy: milky urine which contained enormous amounts of amorphous urates. Uric acid determinations of the blood were normal at that time, and the condition cleared up spontaneously in both cases after sulfathiazole was discontinued.

Postoperative management

Four cases received blood transfusions postoperatively, one of them twice (500 cc. of red blood cells in 500 cc. of normal saline). All four cases receiving sulfathiazole postoperatively, in full doses, complained of the reactions described above. The highest total dose given per patient was 65.6 gm. and the lowest, 20 gm. All cases received vitamins parenterally and hematinics by mouth.

Condition of perineal anus on discharge

All were fully patent. Five showed moderate to slight degrees of protrusion that necessitated further surgery. Three cases had moderate bowel continency; the other six were incontinent and averaged two to three soft stools a day.

Readmissions for further surgery

Three patients were readmitted for further surgery of the perineal anus—one case had excision of the prolapsed portion of the colostomy and, subsequently, a cicatricial stricture of the anus. This patient was again readmitted for plastic surgery after excision of the scar tissue. She will have to return at some other time for repair of a large postoperative ventral hernia. Two other patients had, respectively, excision and cauterization of a redundant portion of the perineal anus. One of these had to be treated further at the dispensary with quinine and urea sclerosing injections; still another case had similar treatment.

Follow up

To date all cases have been followed up—four cases during seventeen months and two others during fifteen months, which constitute the longest follow-up periods. The shortest was during two months, with an average of 12.6 months. A slight contracture of the perineal anus has appeared in all cases but this condition has been treated successfully by dilatations with the gloved finger or with rubber

sounds. A cicatricial ring of fibrosis at the mucocutaneous junction has been noticed in three cases. Two patients have no sphincter control but regulate their bowel movements satisfactorily by means of enemas every two or three days. The male patient is incontinent at times and has to wear a protective pad if he does not use enemas periodically. The remaining nine patients have very satisfactory sphincter control and move their bowels twice a day, daily, or every other day. The gain in weight, strength, and health has been remarkable in all cases.

These follow-up periods have not been sufficiently long to consider the patients as totally cured and only prolonged observation will determine whether recurrences are to occur.

ADDITIONAL CASES

During the first eight months of 1943 and while this study was still in preparation, three more cases were successfully treated by our radical technique. In order that these cases may be included in the analysis of the follow-up period and of the results obtained and so that they may serve as illustration of our present discussion, a brief summary of each is appended hereto.

CASE 10. U. H. 3655. A 38-year-old white female was admitted on February 5, 1943 to the University Hospital, with a history of constipation, tenesmus, painful defecation, ribbon stools containing blood, pus and mucus, irritating rectal discharge, marked flatulence, and a loss of 40 pounds in weight. These symptoms had been present for six years. In addition, general malaise and weakness were apparent and a pararectal abscess had formed a year previous to admission. The patient denied sexual intercourse per rectum and stated that her blood was negative for syphilis. However, a Frei test had proved positive two weeks prior to hospitalization.

A physical examination showed that the patient weighed only 81 pounds. There were noticeable signs of undernourishment. The inguinal lymph glands were enlarged bilaterally; the sigmoid was tender and full of feces. Anoscopic examination revealed a mucocutaneous tag at six o'clock, a toneless sphincter, and a granular, hyperemic mucosa throughout the distal 3 cm. of the rectum. Just proximal to this area the mucosa was ulcerated and bled easily and there was an unyielding stricture of the tubular variety with fibrotic walls.

Significant laboratory findings were a moderate normochromic

anemia, a negative Kahn test, a sedimentation rate of 34 mm. in one hour, and a total serum protein level of 9.80 gm. percent, with 5.37 gm. percent of albumin and 4.53 gm. percent of globulin.

During the preoperative period of twelve days the patient suffered from an acute arthritis of the left knee which subsided spontaneously but which was considered lymphogranulomatous in nature.

The operation was performed on February 17, 1943—the thirteenth day of hospitalization—under a satisfactory continuous spinal anaesthesia in which a total of 300 mgm. of procaine crystals was used. The operation lasted one hour and twenty-five minutes; during its course a 500 cc. blood transfusion was given. Marked perirectal fibrosis and granulomatous masses were encountered; the stricture was tubular in type. An accidental tear of the sigmoidal wall occurred during dissection.

The pathologic report showed an acute and chronic rectosigmoiditis with ulceration and stricture due to lymphogranuloma venereum. The postoperative course was complicated by a paralytic ileus during the first three days; on the fifth, the perineal packing was removed. The end of the sigmoid was gangrenous but it healed uneventfully after the dead tissue sloughed away.

The patient was discharged on the twenty-eighth postoperative day—March 17, 1943—without the ability to control her bowel movements. Three months later and, after periodic rectal dilations at the dispensary, she had to be readmitted because of an acute intestinal obstruction due to gangrene of a loop of the small intestine, apparently caused by constriction of the loop by a fibrous band. Resection was successful and the patient was discharged on May 5, 1943, the seventeenth postoperative day. By July, 1943, her weight had increased to 86 pounds and she was having regular bowel movements. The patient was last seen on September 29, 1943, reporting good bowel control and regular daily defecations. Her weight at the time was 91 pounds.

CASE 11. U. H. 4087. This 24-year-old colored, married female was admitted on May 6, 1943. She gave a history of a gradually increasing constipation of many years' duration, complicated during the previous two months by tenesmus, painful defecation and blood, pus, and mucus in the stools. The patient had been treated for syphilis with salvarsan and bismuth injections.

The physical examination revealed a frail and undernourished young Negro, presenting a scar in the left groin. Proctoscopy showed a spastic sphincter, an indurated, fixed rectal wall, and a

stricture some 4-5 cm. above the anus which barely admitted the tip of the index finger.

Significant laboratory findings were a positive Frei test, a moderate hypochromic anemia, a negative Kahn test, and a total serum protein value of 13 gm. percent, with 6.45 gm. percent of albumin and 6.55 gm. percent of globulin. After three weeks of sulfadiazine therapy (the patient had developed a papular rash when sulfathiazole was tried), laxatives, a low residue diet, and rest, she was operated upon May 26, 1943. A satisfactory continuous spinal anaesthesia was maintained with a total of 200 mgm. of procaine crystals. The operation lasted one and a half hours. The rectal wall and perirectal structures were thickened and indurated. There was a 6-7 cm. tubular stricture some 5 cm. above the anal opening.

The pathologic report read: "Ulceration and stricture due to lymphogranuloma inguinale (?); acute and chronic proctitis and periproctitis; Schistosomiasis mansoni."

The patient's convalescence was an easy one and she was discharged on June 6, 1943, the fourteenth day after operation. By July 20, 1943, she had gained seven pounds in weight and was having two to three well-formed and well-controlled daily stools. There was, however, a slight prolapse of the mucosa which was successfully treated with small quantities of quinine and urea hydrochloride solution in injections. The patient was last seen on October 6, 1943, at which time she reported good bowel movements, with full control, and a total weight gain of 12 pounds.

CASE 12. U. H. 4339. This 40-year-old white widow was admitted July 21, 1943, with a history of constipation, ribbon stools, tenesmus, and painful, bloody, purulent stools of five years' duration. Three years prior to admission she had developed a pararectal abscess. Six months previous to admission she had undergone a Frei test at another hospital and this had given a positive reaction. The patient was then treated with some type of injection and by manual dilations; improvement was slight, however.

The physical examination showed an undernourished middle-aged female, weighing 98 pounds. She had a stricture 3-4 cm. above the anus, barely admitting the finger tip.

Laboratory studies resulted in a positive Frei test, a moderate normochromic anemia, a Kahn 3-3-3, Wassermann 2+, a positive Hinton, and a sedimentation rate of 41 mm. per hour, with 10.3 gm. percent of total serum proteins, 4.3 gm. percent of albumin, and 6 gm. percent of globulin.

The patient received sulfathiazole for five days and was operated upon August 8, 1943, the thirteenth day of hospitalization. Continuous spinal anaesthesia was used and a total of 400 mgm. of procaine employed. The operation lasted two hours and disclosed marked fibrosis of the perirectal tissues and a thickening of the rectal wall which was adherent to the posterior wall of the vagina. The rectal stricture was very extensive, measuring about 20 cm. in length. The wall of the rectum was accidentally torn in one place during dissection.

Postoperatively, there developed a severe paralytic ileus that lasted twelve days; it was treated at first with the Wangenstein suction and later with the Miller-Abbott tube. Infusions of 10 percent glucose in normal saline were given daily. The patient responded well to this treatment and was discharged August 8, 1943, on the twentieth day after operation. At the time of release her bowel movements were partially under control.

Quinine and urea injections, administered later in the dispensary, controlled a mild degree of mucosal prolapse of the perineal anus. On September 14, 1943, a postoperative diastasis recti was observed. On October 13, 1943, the patient reported good bowel control and regular daily stools.

SUMMARY

1. Various methods of surgical approach to the problem of lymphogranulomatous rectal stricture are discussed and the literature on the subject is reviewed and evaluated.
2. A modification of the Babcock abdominoperineal resection is presented and proposed for the radical cure of stricture of the rectum due to lymphogranuloma venereum, based on twelve cases with no mortality.
3. A detailed analysis of nine of these cases and a summary of three others is herein presented.