

Lucite had already been found to produce little tissue or skin reaction,³ and Lucite face plates for application to the skin have been in use for nearly three years at University of Minnesota Hospitals. Over a period of many months, Vinylite bags* have been attached to these plates, but have been discarded because the material becomes discolored and stiff within a few days. Early in 1949 the decision was made to use polyethylene bags attached to a deeply grooved flange on the face plate by over-wrapping with rubber binders. This combination has performed well to date, although the tensile strength is less than desired.

The detail of the present bag is indicated in the accompanying illustration. Opaque white bags are being obtained, as less offensive from an esthetic point of view than transparent containers.

Rutzen cement does not adhere well to the Lucite face plate. Ordinary Dermatome† cement is spread on the face plate, and Rutzen skin cement on the skin. After drying for three minutes, the bag is applied precisely, and adhesion is as secure as is the case with the standard Koenig-Rutzen bag.

A third problem has arisen in the apparent sensitivity of the skin of a few patients to the rubber cement. A method for use of a special pressure sensitive adhesive tape‡ has been under study in collaboration with the Minnesota Mining and Manufacturing Company, which may eventually overcome this problem as well.

At the present time, twelve patients have worn such appliances as have been described and have contributed ideas and criticisms which have brought the apparatus to the present state. Steps are planned to make such an apparatus generally available in the near future.§

SUMMARY

1. Methods to assure sound healing and healthy skin about ileostomies are indicated.
2. A bag is described which is valuable in those ileostomy patients whose discharge destroys the rubber of the Koenig-Rutzen bag.

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*Manufactured as hot water bottles by the Alden Wonder Plastics Company, Minneapolis, Minn.

†Obtained from V. Mueller & Company, Chicago, Ill. Manufactured by Padgett-Hood Assemblage Company, Kansas City, Mo.

‡Through the courtesy of L. W. Lehr and Bert Auger, "Scotch" Tape Division, Minnesota Mining and Manufacturing Company, St. Paul, Minn.

§A Lucite and Polythene bag of very different design is marketed by Saul Saetz of Burlington, Vt. It came to our attention some months after adoption of our present design. The bag does not satisfy the criteria outlined in this paper in that it is bulky and insecure. After exchange of information, he also is evaluating a Minnesota Mining and Manufacturing Company type of cement.

A NEW SURGICAL TREATMENT FOR HIRSCHSPRUNG'S DISEASE

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THE pathology in Hirschsprung's disease is generally considered to be limited to the dilated portion of the colon. Recently, evidence has accumulated raising questions in connection with this theory and supporting the concept of a functional obstruction in the rectosigmoid which produces secondarily the hypertrophied and dilated colon. This evidence is as follows:

1. Colostomy relieves all of the symptoms of Hirschsprung's disease and in three to six months the dilated colon becomes normal in size and much of the hypertrophy disappears.¹ If the primary pathology were in the dilated colon, no relief should be gained from a colostomy. One would expect relief from a colostomy if the basic pathology were a low partial colonic obstruction (Fig. 1). Closure of the colostomy in patients with Hirschsprung's disease produces a recurrence of the disease in three to four months.

2. Barium enema studies on patients with Hirschsprung's disease provide additional evidence of pathology in the rectosigmoid.² Careful examination of the colon demonstrates that the distal dilated sigmoid has a funnel-shaped outline and that the rectosigmoid is narrow and irregular (Fig. 2). Frequently, this lesion is difficult to outline either by fluoroscopy or by roentgen films. It can best be demonstrated by the injection of a small amount of barium, with the patient in the oblique position. Most important, the dilated sigmoid must be manipulated out of the pelvis to permit a clear view of the rectosigmoid.

3. The motility of the colon in Hirschsprung's disease has been studied and it has been found that the dilated colon has normal, integrated peristalsis. Furthermore, it has been demonstrated that patients with Hirschsprung's disease have no peristalsis in the rectosigmoid, in contrast to normal individuals. It is postulated that this absence of peristalsis results in a partial functional obstruction.³

4. The resection of this functionally deficient segment of colon by a special technique in fifty-two patients has relieved their symptoms.

5. Histologic study of the specimens of colon surgically removed from patients has demonstrated the absence of Auerbach's plexus in the rectosigmoid.³ Similar findings have been reported from other studies.⁴⁻⁸ According to Cannon, peristalsis cannot take place in bowel in which Auerbach's plexuses have been destroyed.⁹

It would seem on the basis of this evidence that the fundamental lesion in Hirschsprung's disease is a congenital one, consisting of a failure of development of Auerbach's plexuses in the rectum and rectosigmoid. Auerbach's plexuses contain the ganglia of the parasympathetic supply to the bowel. Most of the

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colon, including the descending portion, probably receives its parasympathetic supply from the vagus. The distal end of the colon receives its parasympathetic supply from the pelvic nerves. Failure of the pelvic parasympathetic supply to develop may be the explanation of the location of the lesion in Hirschsprung's

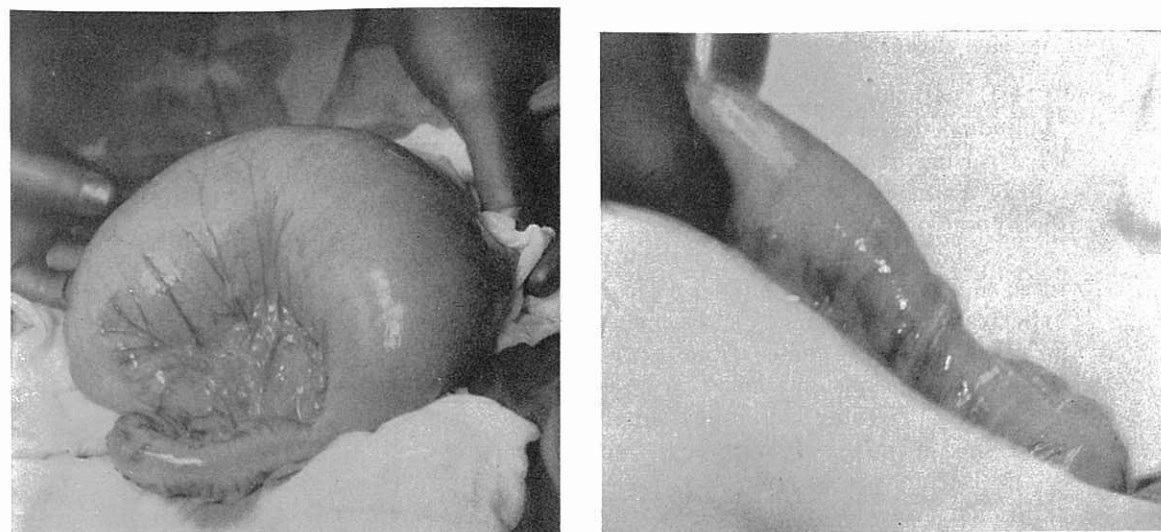


Fig. 1.—A, Photograph of sigmoid during course of colostomy. B, Photograph of same bowel four months later at time of resection.

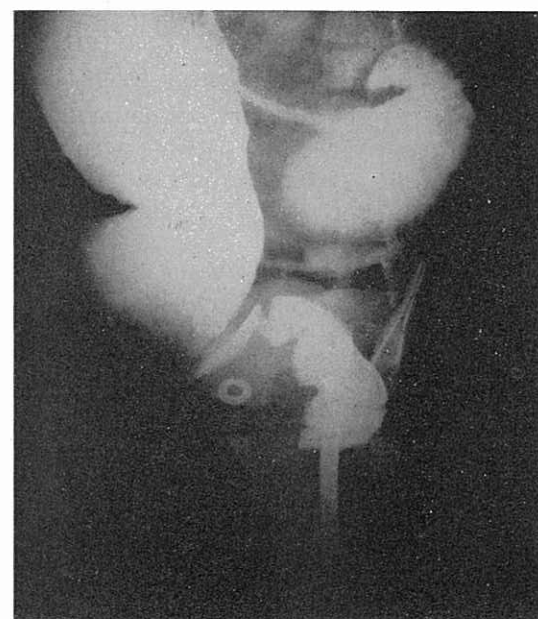


Fig. 2.—Barium enema showing narrowing in rectosigmoid and dilatation of the sigmoid.

disease. The dilated colon in Hirschsprung's disease contains normal nerve structure. This evidence indicates that therapeutic efforts should be directed to the rectosigmoid and rectum and not to the dilated colon.

The selection of patients with megacolon suitable for surgical therapy depends on the demonstration of a narrowing of the rectosigmoid by barium enema studies. Patients with a colon dilated down to the anus do not have Hirschsprung's disease, according to our concept, and will not benefit from resection of the rectosigmoid and rectum.

Out of the first twenty-two patients with Hirschsprung's disease treated surgically, sixteen had a colostomy performed as a preliminary measure. As our experience has grown, fewer colostomies have been performed. All colostomies are now performed in the transverse colon and are of the simple loop type.

In only six of the last thirty patients has a colostomy been considered necessary. In five of these cases the infants were extremely ill and primary resection was considered inadvisable. In one patient it was impossible to prepare the bowel for a primary resection.

Preoperative preparation, which begins four weeks before admission to the hospital, consists of a low residue diet, two ounces of mineral oil daily, and one to two enemas each day. When the patient is admitted for operation, a large rectal tube is inserted for a sufficient distance to permit the end to be in the dilated colon. It is taped in place and colonic washings are then performed three times a day. Usually the tube can be positioned quite readily. However, in one patient proctoscopy under general anesthesia was performed to position the tube. Even this procedure failed in a second patient and this accounts for one recent colostomy. After the rectal tube has been in place twenty-four to forty-eight hours the patient's abdomen is scaphoid and he is ready for primary resection.

Most patients with Hirschsprung's disease have a defective urinary bladder. Of twenty patients in our group studied by cystometrogram preoperatively all except one had hypotonic bladders. The bladder capacity of these patients before any detrusor contractions occurred was considered far above average. This is probably further evidence of a defective pelvic parasympathetic supply. The presence of a defective bladder is extremely important from a technical standpoint for slight damage to the bladder innervation during operation may result in urinary retention.

The experimental work on which our special technique for resection of the rectosigmoid and rectum is based has been described previously.¹ The patient is placed in a lithotomy position which permits access to both the abdomen and the perineum (Fig. 3). The draping is so arranged that at the completion of the first stage of the operation the removal of a heavy double sheet from the legs of the patient exposes the perineum without disturbing the abdominal drapes.

A suprapubic incision slightly to the left of the midline is used. The sigmoid loop is delivered and usually the transition from dilated to narrow

colon occurs at the junction of the sigmoid and rectosigmoid. However, in our series, varying portions of the sigmoid have been involved and in three patients all of it was narrow. In two patients, all of the colon from the left one-half of the transverse to the anus was involved. In three patients only the rectum was involved.

Pathologic studies have demonstrated the absence of Auerbach's plexus to extend some distance into the dilated bowel. Therefore, the proximal point of resection should be at least 12 cm. proximal to the point of narrowing to insure removal of all of the lesion.



Fig. 3.—Position of patient on operating table.

The next step is to free the colon proximal to the resection so that it can be brought to the perineum. There is always adequate bowel; the limiting factors are the blood vessels. To free the sigmoid sufficiently it is necessary to divide the sigmoidal branches at their origin (Fig. 4). An arcade system is thus preserved which supplies the circulation of the bowel adequately. On six occasions we have taken down the splenic flexure and divided the left colic artery, thus permitting resection of most of the descending colon and sigmoid. In two patients a left hemicolectomy was performed with division of the middle colic artery. So far there has been no failure of the blood supply to the bowel proximal to the resection.

The dissection of the rectosigmoid and rectum is then performed right down to the internal sphincter. This must be done with great care or there may be permanent damage to the innervation of the bladder. It must be remembered that patients with Hirschsprung's disease have a damaged bladder mechanism. Therefore, the margin of safety is small in these patients and the dissection must be performed carefully.

It is our practice to remove a segment of bowel to facilitate pulling the distal part through the anus. Using the von Petz clamp the bowel is divided at the previously selected point of resection and again distal to this at the juncture of the sigmoid and rectosigmoid (Fig. 4). The remaining stump of sigmoid can readily be pulled through the anus.

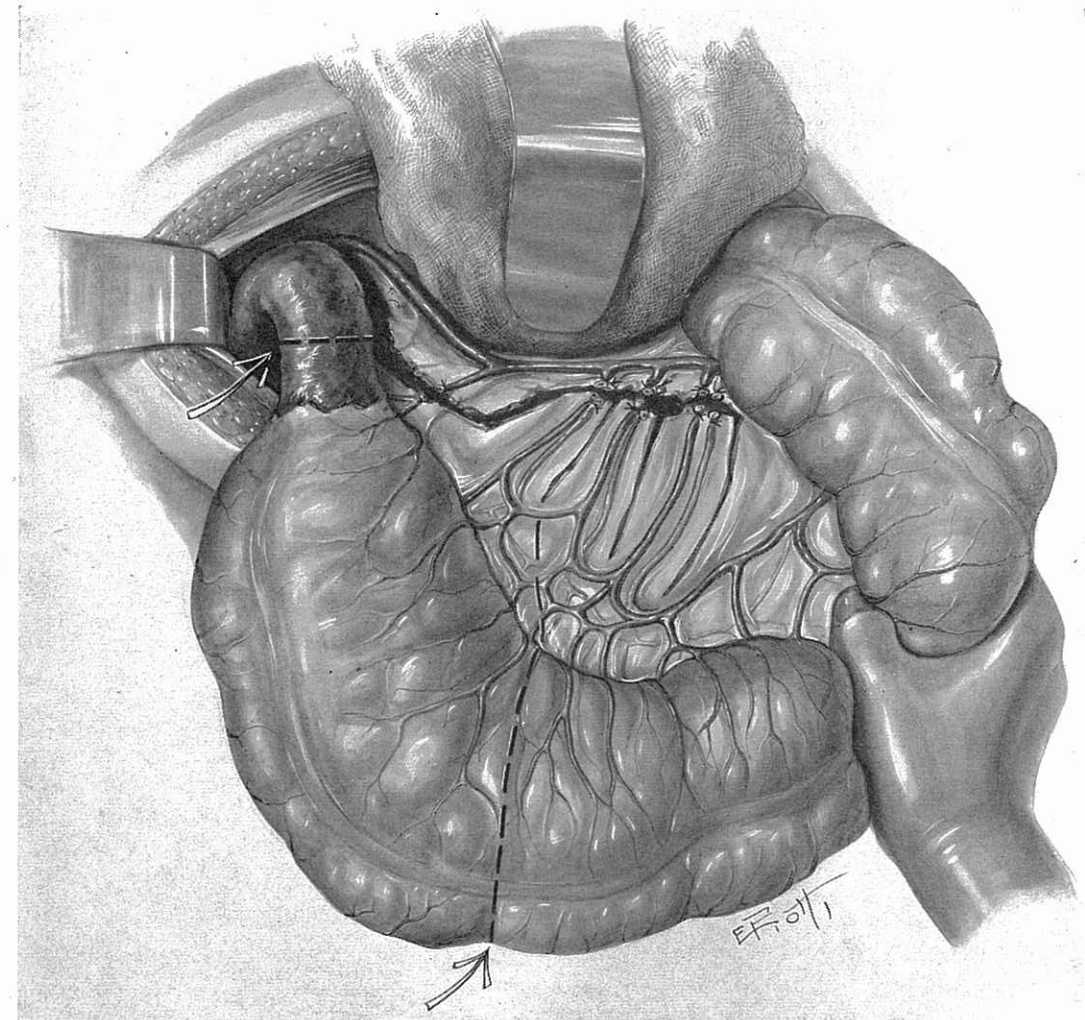


Fig. 4.—Drawing showing the division of three sigmoidal branches. The broken lines indicate the segment of bowel which is resected. This facilitates pulling the rectosigmoid out through the anus.

At this stage of the procedure the operative team is divided into two groups and the drape over the legs is removed, exposing the perineum. The sphincter is dilated and a long clamp inserted through the anus up to the closed end of the rectosigmoid (Fig. 5). The end is grasped and pulled down through the anus, turning the bowel inside out. This segment, protruding from the

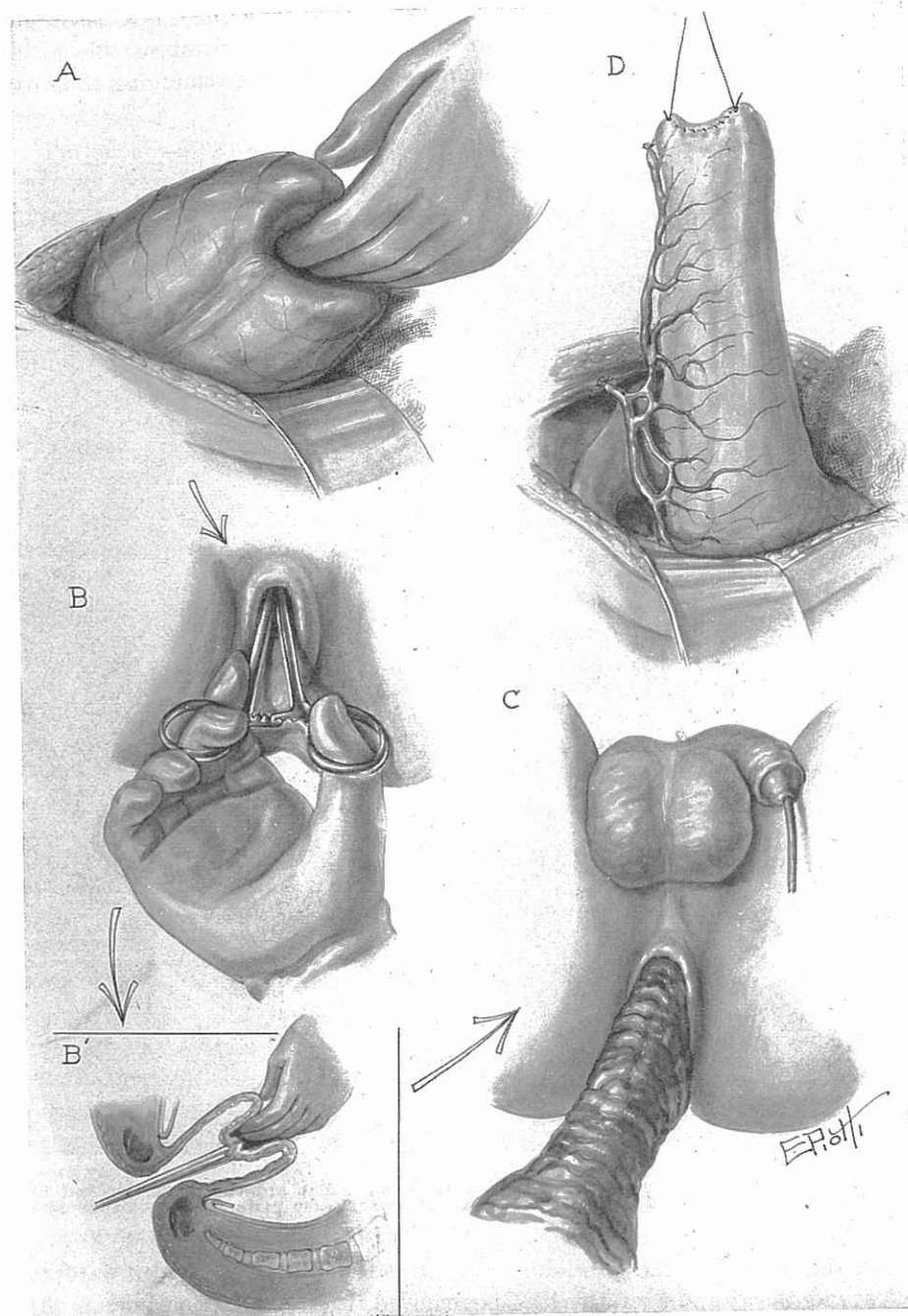


Fig. 5.—A and B, A clamp is inserted through the anus and the closed end of the rectosigmoid is grasped. B', The bowel is then pulled out through the anus. C, The segment now protrudes with the mucosa on the outside. D, The proximal bowel has two sutures placed to facilitate pulling this segment through.

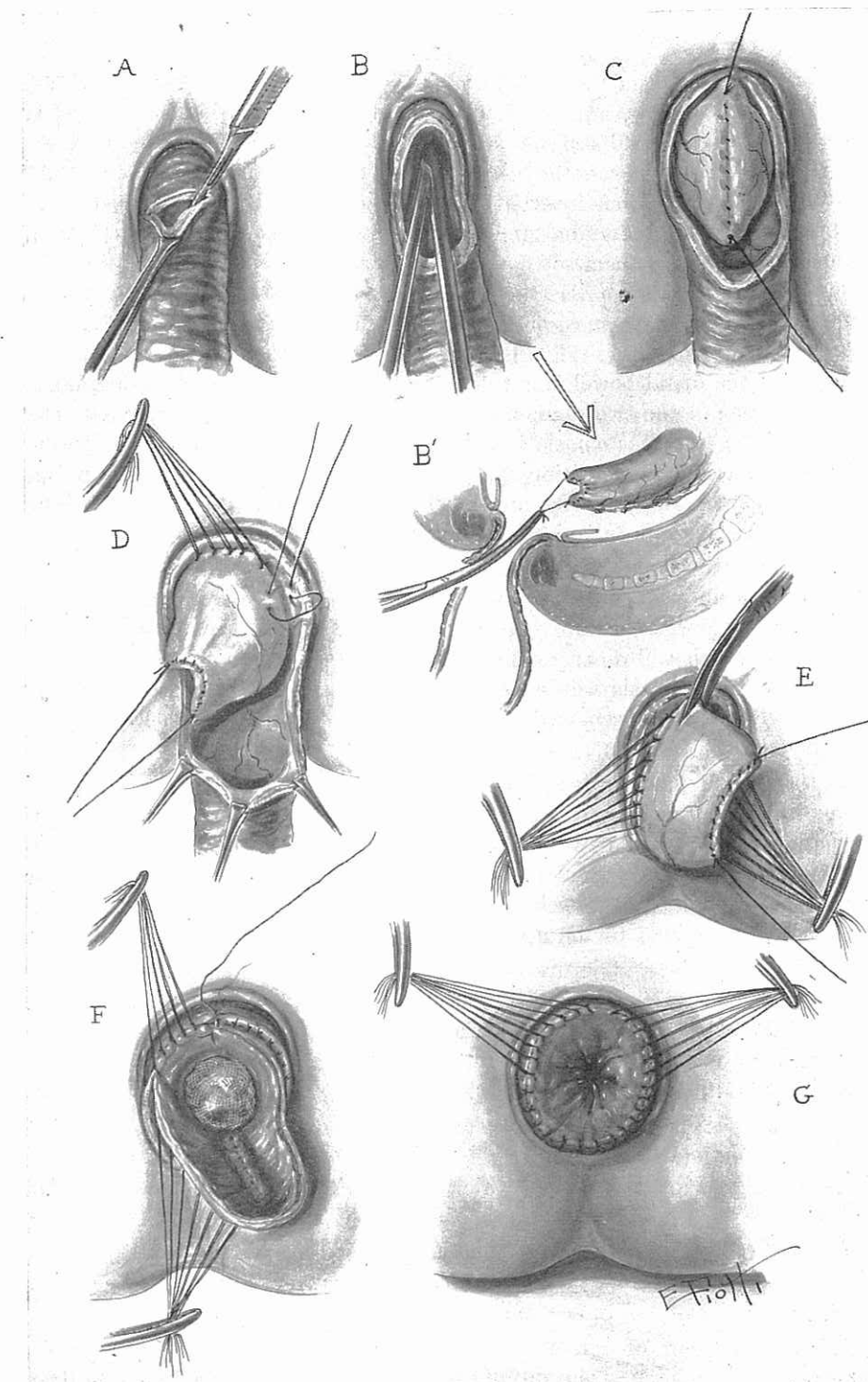


Fig. 6.—A, After the bowel is prepared and drapes placed, a cut is made 2 or 3 cm. from the mucocutaneous margin. B, A clamp is inserted through this orifice and the sutures on the proximal segment grasped and the bowel pulled through as in C. D, Interrupted silk sutures are used to approximate the muscular coats. E, The proximal bowel is opened. F, The mucosae are approximated with catgut sutures. G, Completion of the anastomosis. After all the sutures are cut, the anastomotic line will retract up through the anus.

anus with the mucosa on the outside, and the perineum are scrubbed with ten sponges soaked in 1:1000 aqueous Zephiran. The perineum is then draped and a table of instruments and supplies set up for the team working at the perineum. A cut is made halfway across the bowel $2\frac{1}{2}$ cm. from the cutaneous margin (Fig. 6). A long clamp is then inserted through this opening into the pelvis, and the operator at the abdominal incision, having attached two sutures to the proximal bowel, places them so that they are grasped by the clamp. It is then possible to pull the proximal segment down through the pelvis and out through the distal segment. The anastomosis is made, using first a row of interrupted silk sutures to approximate the muscular coats. As these sutures are placed the remainder of the distal bowel is cut across. The proximal bowel is then opened and the mucosae of the two segments are approximated with interrupted catgut sutures. When the anastomosis is completed it is allowed to retract through the anus and assumes a position 2 to 3 cm. from the mucocutaneous margin. While the anastomosis is being made below, the team at the abdominal field reconstruct the pelvic floor and close the abdominal wound.

The alternative to this pull-through type of anastomosis would be the performance of an anterior resection—a procedure which has several disadvantages in Hirschsprung's disease. In the first place, in the pull-through operation we remove all of the bowel down to the internal sphincter. In the small pelvis of a child a low anastomosis would be difficult to perform and therefore some of the lesion would not be removed. Two recurrences following such procedures have come to my attention.

In the second place, using the pull-through technique we can perform resection on most patients without making a preliminary colostomy because the discrepancy in size of the two segments is minimized by the dilatation of the distal segment as the proximal segment is pulled through. If the anterior resection is contemplated, a preliminary colostomy is required to permit the dilated bowel to return to the approximate size of the distal narrow segment.

In the third place, stricture at the suture line is less likely to occur if a pull-through anastomosis is made. The dilated colon permits a large stoma at the suture line and we have had only one slight narrowing of the suture line in our primary resections.

In the fourth place, with the pull-through procedure the exposure during the anastomosis is excellent so that leaks and infection are rare. There has been one case of infection in our group. This was not a routine case, as it was complicated by a previous colostomy which limited the length of bowel available and hindered the establishment of a satisfactory anastomosis.

In the fifth place, our method reduces the operating time and permits us to perform resection on sick patients who would require a preliminary colostomy were an anterior resection to be used.

The management of the urinary bladder is the most important part of the postoperative regime. The inlying bladder catheter is placed on free drainage for four days. Tidal irrigation is then used until the tenth postoperative day, when the catheter is removed. We have had only two urinary complications.

The first was infection one week after discharge, which responded to penicillin. The second was difficulty in emptying the bladder. This symptom cleared up two weeks after the catheter was removed. Furmethide was used during this period and seemed to be of value.

Postoperatively, the patients are given a regular diet without laxatives or mineral oil. Occasionally, during the early postoperative period, before peristalsis is present in the colon, it may be necessary to insert a rectal tube two or three times a day to help decompress the patient.

Following discharge from the hospital, the patients have a diarrhea lasting sometimes from three to six weeks. Gradually they settle down to a system of one to two bowel movements a day.

One slight stricture has occurred in this series of thirty-one patients with primary resection and it was probably due to tension on the suture line. It was corrected by a short course of digital dilatations. There have been three other strictures, all in patients with preliminary colostomies. The colon had shrunk to 2 cm. or less in diameter and the stoma at the anastomosis was correspondingly small. Later, as the bowel dilated to normal size, the suture line remained fixed in size and required dilatation. This experience stresses the necessity of allowing only three to six weeks between colostomy and resection. If the interval is much longer, the bowel becomes so small that a narrowing at the suture line is inevitable. These patients have also done well on digital dilatation.

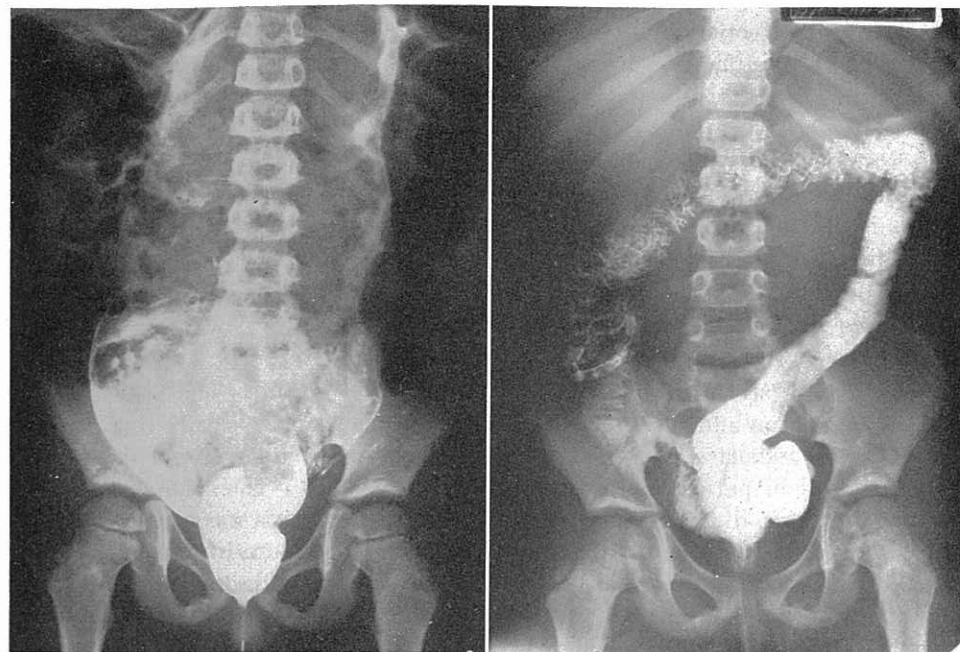
The most frequent complication has been acute gastroenteritis, which these patients tolerate poorly. This difficulty has occurred in nine patients, most of them under 2 years of age. Three children were seriously ill, with vomiting, distention, and diarrhea. We have found that the best treatment has been the insertion of a rectal tube and colonic washing. In two to three days the patients have recovered, the abdominal distention has disappeared, and bowel movements have become formed and regular.

We have had one failure in a group of fifty-two patients operated upon. This was a baby 2 weeks of age who did not have a typical lesion at operation. The sigmoid and rectosigmoid were resected and no ganglion cells were found in the resected bowel. Abdominal distention and constipation recurred two months after operation. By x-ray examination the lesion was found to extend to the splenic flexure. A second resection has recently been performed on this patient.

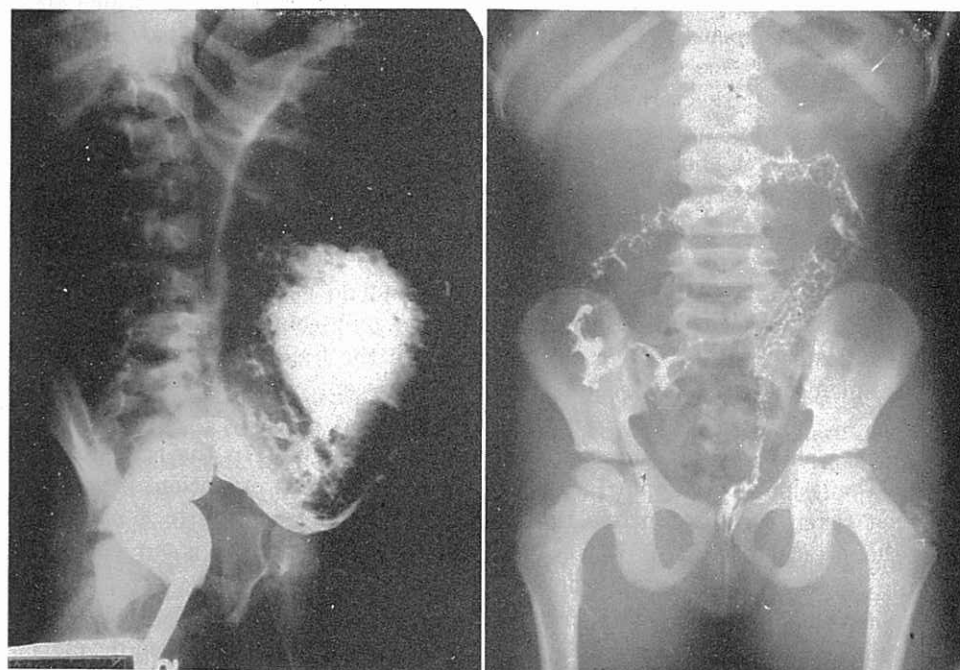
It is well to avoid operating on infants if possible. In the first place, in babies the lesion is not typical by x-ray examination or at operation. Dilatation has not taken place to outline the extent of the lesion. At 6 to 8 months of age the typical dilatation has occurred; consequently, there is no danger of failing to remove all of the lesion.

There has been one postoperative death which occurred in the case of a 3-week-old infant following a primary resection. Intestinal obstruction developed, which was corrected. However, eventually the baby died of agranulocytosis.

The remaining patients have been completely relieved of their complaints. The longest follow-up is almost three years. All of the patients have normal



A.



B.

Fig. 7.—A, Preoperative barium enema and a checkup three months after operation. B, Preoperative barium enema and a repeat six months after operation.

sphincter control. They are all on normal diets, with no mineral oil or other laxatives, and have one to two bowel movements a day. All of them have gained weight and are in good general health.

Our results with this form of treatment have been corroborated by Stephens.⁸ After visiting us and observing our method he returned to England and subsequently operated upon twenty-eight patients using our technique. There were two postoperative deaths. In the remaining patients he reported good clinical results.

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DISCUSSION

DR. K. GRIMSON.—Dr. Swenson certainly deserves a good word for his excellent studies and congratulations on his large series. He has well demonstrated that like colostomy, the pull-through procedure will effect reduction in the size of the colon. His concept of the etiology is probably correct, but during seven years of study of approximately thirty megacolon patients we have not been able to prove it.

The normal-sized but abnormally functioning lower sigmoid and rectum, according to our balloons and other observations, is not functionless but rather, though working abnormally, can contract and particularly dilate.

Four years ago, we reported our belief that surgery was generally unnecessary for the sigmoid-achalasia type of Hirschsprung's disease or megacolon. In spite of Dr. Swenson's operations and reports our additional experiences have strengthened this belief. Many of our patients had dilatation of the descending and sigmoid colon as great as those presented by slides this afternoon. However, with an improved medical program of treatment these children were carefully carried through a few troublesome years following which symptoms usually subsided and bowel movements occurred regularly in later life.

Only in three patients who did not have the sigmoid-achalasia type of megacolon but rather had enormous enlargement of the right, transverse, and upper left colon above a normal-appearing rectum and lower sigmoid was primary operation considered necessary and performed. In these, as reported, we used a colectomy and ileosigmoidostomy to direct the liquid content of the terminal ileum into a segment of the distal and not dilated sigmoid. The stump of sigmoid left should be short.

In only one additional patient during the study period in this series was surgery necessary. For this child a pull-through procedure was done because of previous surgery starting with a colostomy shortly after birth. The result has been only moderately satisfactory, some dilatation and some obstipation persisting.

I certainly feel that Dr. Swenson has given a very nice demonstration and that his studies and experiences should aid us in determining cause and proper treatment of megacolon.

DR. M. RAVITCH.—This is a large, beautiful series and the results could hardly be improved upon. But there still remain some facts to be explained about Hirschsprung's disease and its histopathology.

When Bodian and Stephens revived the idea of the absence of the ganglion cells in the segment which Dr. Swenson had felt was the site of the disease, they began operating—Stephens did, after Dr. Swenson—and have now a considerable series. It is their opinion that uniformly, the ganglion cells are absent in the narrow distal segment, and at times this absence extends several centimeters into the dilated section.

I have studied their sections in a considerable number of cases and must say the findings they describe in the sections they have can be seen by anyone.

In a very small number of cases of our own, we have not been able to convince ourselves with certainty that the absence of ganglion cells in the narrow segment is invariable, and is not perhaps some type of artifact due to technique or varying thickness of the bowel or contraction or twisting.

I would like to ask Dr. Swenson whether he still feels as he stated in a recent publication that those histologic abnormalities in his experience are not invariable.

The other thing which is difficult for us to explain is the fact that we have had clinically quite good results from resection of the left colon, much as Dr. Bell reported some years ago, beginning to the left of the middle colic artery and going down well into the pelvis. If there is a narrow segment, we go down into it; if there is not a narrow segment, we do a conveniently low, safe, aseptic, closed anastomosis.

Several months ago we operated upon a girl of 14 who had never had a spontaneous stool. She had the most elegant narrow segment imaginable going up almost to the splenic flexure. Knowing that Dr. Stephens and Dr. Swenson would have resected the narrow segment and left the dilated segment, we resected the dilated and left the narrow, anastomosing the ileum to the descending colon. She began having spontaneous stools the second or third day and has had them ever since, although she had never had them before. She has not had an enema for the three or four months since the operation.

I wonder if it is possible that some of the results achieved from this ingenious operation are due to straightening out the redundant bowel. The two patients, for instance, in whom a rectal tube could not be passed might very well have had a chronic volvulus. In several cases we have seen the bowel twisted on itself as it lay across the junction of the narrow and dilated bowel.

We have tried this pull-through procedure. It is technically easy and perfectly satisfactory as an operation, but thus far, in a much smaller series than Dr. Swenson's, we prefer the results with resection of the left colon.

DR. G. HUMPHREYS.—I want to join in stating that I think the concept of Hirschsprung's disease as being due to physiologic disturbance in the small segment is a tremendous contribution in treating this disease. Dr. Hiatt at Presbyterian Hospital started to work on this about four years ago and has paralleled the work of the Boston group very completely.

In our balloon studies, the function has been similar to what Dr. Grimson just described, that is, rather than finding a functionless segment we find one that does contract but contracts in a mass rather than in a peristaltic way.

In other words, if one puts three balloons in the distal small segment rather than having two in the dilated and one in the small, one finds that the wave which comes down to that segment then causes a mass contraction which in effect causes an obstruction.

Now the histology has puzzled all of us, also. I think some of the discussion is confused by a statement that Auerbach's plexus is absent. That segment is not devoid of nerves. There are plenty of nerves in it as far as we can see histologically. It is apparently

lacking in ganglion cells and that seems to be the source of the confusion, since there is no good basis for judging what the normal number of ganglion cells is.

It is our conception that this physiologic imbalance may vary a good deal in different patients. Some may have a very sharply limited area and others may have a line of demarcation which is much less distinct, so that they have a more or less funnel-shaped end to their dilated colon; and that where this area of deficient but not completely absent innervation comes down to the rectum, one has the group in whom the dilated bowel extends right to the rectum or may actually overcome the rectal sphincter tone, and so apparently result in the spontaneous remission later in life that Dr. Grimson speaks of.

It certainly has been a great help in treating these patients to have this concept. Dr. Hiatt's operation is a pull-through operation very much like Dr. Swenson's, and we have had equally good results—again in a much smaller series.

DR. C. E. KOOP.—I shall make my remarks very brief. About three years ago we gathered together sixteen patients who had megacolon and I was about to publish a paper stating that chronic volvulus was the etiological factor. At that time I knew of Dr. Swenson's work and set about reviewing our own cases, and have proved to my own satisfaction that he is correct. Our balloon studies have paralleled his. Our clinical impressions have paralleled his, and histologically we see the changes in the myenteric plexus in about 80 per cent of patients.

I think the pertinent thing about my comment is that in those sixteen patients we did anterior resections as low as we possibly could. They all did well for fourteen months. After fourteen months three of them had recurrence of their symptoms with changes in the barium enema. Of those three patients I have operated again on two of them and have done the pull-through procedure.

In reviewing the previously excised bowels and the present excision of bowel, the changes in the myenteric plexus support Dr. Swenson's thesis.

Since that time we have carried out an additional fourteen pull-through procedures with no failures.

DR. O. SWENSON (Closing).—The pathology of Hirschsprung's disease has been carefully studied and there is a unanimity of opinion in the published reports. Dalla Valle, in 1920, was the first to report absence of Auerbach's plexus in the distal colon. Roberts and Kernohan reported the same findings in Hirschsprung's disease in 1938. Tiffin, Chandler and Faber, in the *American Journal of Diseases of Children* in 1940, and Zuelzer and Wilson, in the same journal in 1948, reported absence of Auerbach's plexus in congenital megacolon.

Cannon demonstrated experimentally that destruction of Auerbach's plexus resulted in absence of peristalsis. This agrees completely with our motility studies. In the segments of bowel where there was an absence of peristalsis by motility studies there were no Auerbach's plexuses in the microscopic sections.

In following patients with anterior resection or resection of the dilated colon alone, three or four months' follow-up is not sufficient. This disease is the result of a functional partial obstruction and therefore in some cases recurrence does not occur until six to eight months postoperatively.

We have followed up some of our patients almost three years now and they continue to be perfectly well.