PERIANAL FISTULAE AS A COMPLICATION OF REGIONAL ILEITIS

ABRAHAM PENNER, M.D.
AND
BURRILL B. CROHN, M.D.
NEW YORK, N. Y.
FROM THE MEDICAL SERVICES, MT. SINAI HOSPITAL, NEW YORK CITY, N. Y.

SINUS tracts and fistulae of various kinds constitute one of the most constant manifestations of ileitis; they originate in the distal ileum which then constitutes the proximal end of the tract. The fistulae terminate on a surface of the body, epidermal or endodermal. The most common site of termination is on the abdominal wall, the fistulous tract burrowing through the scar of a previous operation (usually an appendicectomy). Where no operative scar is present on the abdominal wall, we have rarely seen fistulae. Other common sites of termination are: Segments of the colon, particularly the cecum, the ascending colon and the ubiquitous sigmoid loop of the pelvic colon; vagina, rectum or ureters.

Of late, in addition, we have received the oral description of a case of ileitis whose earliest and first manifestation was a fecal fistula, making its exit as a perinephric abscess in the right lumbar paravertebral area (Dr. A. Snapper of Amsterdam, Holland). Another case seen by one of us (B.B.C.) presented four lumbar fistulae placed in linear arrangement vertically in the right lumbar area, accompanied by two inguinal fistulae, all of them discharging feces. In neither of these lumbar cases were intestinal symptoms noted, nor was there any suspicion of an intestinal disease until the fecal content of the fistulous abscesses became apparent.

One of the most common types of fistulae complicating ileitis, however, remains to be described; namely, the perianal fistulae. We were not at first conscious of the fact that anal and perianal fistulae constitute a frequent complication of the condition. But with more careful observation we have noted that eight out of 50 analyzed cases of regional ileitis exhibited anal fistulae as a complication, an incidence of 14 per cent. Probably this figure represents much too modest an estimate, for fistula-in-ano, unless questioned for in taking a clinical history, may easily be overlooked, and unless searched for in a complete physical examination, may be completely missed.

It is remarkable to note how frequently the fistula-in-ano was the first clinical manifestation of ileitis, even preceding consciousness of disturbed intestinal function. Such fistulae are usually local phenomena originating, presumably, in a crypt of Morgagni, infected by the contaminated ileal con-

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tents transported to this spot; the tract makes its exit either in the anal region itself, commonly between or through external hemorrhoids, at the anocutaneous margin, on one of the buttocks, in the perineum, or, in women, as a rectovaginal fistula. They may be single or multiple; they may apparently close up for a time, but almost invariably reopen and leak a thin serous, purulent or fecopurulent discharge. They persist as long as the ileitis persists and is active; they close spontaneously when the affected small intestine is resected and may heal when a short-circuiting operation is successful. The rectovaginal fistula is the most persistent and requires surgical removal by dissection, a procedure which is often not successful and frequently must be repeated.

It is of course recognized that fistula-in-ano is a common disease associated with many diarrheal conditions of various origins. On the other hand, simple fistula-in-ano most frequently is a purely local manifestation, independent of any previous or associated disease, or accompanying simple constipation; the nonspecific fistula-in-ano is the type most commonly seen. Tuberculosis, while reputedly the cause of most of these fistulous tracts, is rarely found and is actually an infrequent offender, occurring in only about 1 per cent of instances of fistula-in-ano. Nonspecific ulcerative colitis is commonly complicated by perianal fistulae, according to Bargen, being present in 26 out of 697 instances, or 3.7 per cent of cases of ulcerative colitis. Probably any diarrhea, caused by inflammatory disease of the intestine, may be and is, complicated by fistula-in-ano, no matter how distant the source of the diseased segment. Submucosal infection in the crypts of Morgagni forms a low-grade abscess which breaks inward to the rectal mucosa and outward to the buttock or perianal region.

Noninfectious diarrheas do not produce perianal fistulae. We have never seen gastrogenous, neurogenic or allergic diarrhea, no matter how severe, complicated by fistula-in-ano.

The majority of perineal fistulae complicating ileitis are of local origin in the crypts of Morgagni. We have good reason, however, to suspect that there exist instances of *direct* fistulization from ileum to rectum or to perirectal spaces and then continuously downward to the perineum. The long fistulae originating from the terminal ileum are usually not direct in their course. They are very tortuous and conduct intestinal contents to the perineum. They become secondarily infected, particularly when they head downward and approach the pelvis. Low-grade pelvic abscesses frequently form; the infection seeps or spreads downward into the perirectal or ischiorectal spaces forming a perirectal abscess to the right of the rectal wall as it traverses the pelvis in its extraperitoneal course. This abscess continues to burrow downward and inward to point into the rectal cavity or somewhere near the anus. It is difficult to prove this point as we have not been able to follow such a fistulous tract throughout its continuity. But we have been able to trace, roentgenologically, the fistulous tract from ileum to pararectal abscess; again, in particular instances, we have been able to visualize an oc-

casional peri-anal external fistula, by injecting lipiodol or sodium iodide into its tract and, by roentgenography, prove that its course carries it upward and toward the ampulla of the rectum. Appended are three rather typical case histories which illustrate the existence and course of such "long" fistulous tracts.

CASE REPORTS

Case 1.—We are indebted to Dr. Joseph Tomarkin of Cleveland, Ohio, and Dr. A. A. Berg for data on this case. Hosp. No. 399880: S. G., white, male, age 26, had been complaining of intermittent attacks of abdominal pain for one and one-half years. This pain localized in the center of the abdomen but was not associated with nausea or vomiting. Examination revealed tenderness over McBurney's point accompanied by definite spasticity of the right rectus abdominis. There was also tenderness in the right side of the pelvis on rectal examination. No masses were felt. Temperature was 100° F., leukocyte count, 16,400. Urine analysis was negative. *Preoperative Diagnosis:* Acute appendicitis.

Primary Operation.—The appendix was found to be much enlarged and appeared gangrenous. It was situated in the mesenteric fold of the ileum and presented a small pocket of pus at its tip. The surgeon noted some redness and thickening of the ileum, contiguous to the appendix and extending for a distance of two and one-half to three inches along the terminal loop of the ileum. The appendix was removed and the abdomen was closed with drainage. The patient made an uneventful recovery. *Pathologic Diagnosis:* "Chronic catarrhal and subacute appendicitis and periappendicitis."

He was readmitted to the same hospital in Cleveland, Ohio, two and one-half months later, because of the recurrence of vague epigastric and umbilical pain. There had been no nausea or vomiting and his bowels were regular. Temperature, normal. On this occasion a mass was palpable by rectum; due to this a diagnosis of regional ileitis was suggested. Roentgenologic examination, however, showed no deformities in the ileum or cecum. As a result he was treated conservatively and when, after a few days, the mass, which had been felt by rectum, diminished in size, the patient was discharged.

One month later, he developed colicky pain in the left lower quadrant. There was no nausea or vomiting, diarrhea or constipation. He was running a subfebrile course and had lost a good deal of weight. Examination per rectum revealed a large mass. This was explored with a needle inserted through the lateral rectal wall, and pus was obtained. The abscess cavity was, therefore, drained through the rectum. Biopsy of some tissue from the wall of the abscess was reported as having an acute inflammatory reaction. After some improvement, the fever and pain recurred and the abscess cavity was again drained. About two ounces of pus were obtained and he began to improve rapidly. One month later, he was again readmitted to the same institution, suffering with marked diarrhea and a total loss of 75 pounds in weight. His stools were bloody and defecation was preceded by abdominal cramps. Roentgenologic examination of the small intestine, at this time, showed a typical ileitis with internal fistula formation. He was transfused and transferred to the Mount Sinai Hospital in New York City.

Secondary Operation.—October 30, 1936: He was explored by Dr. A. A. Berg, and was subjected to an ileocecal resection with ileo-ascending colostomy and excision of fistulae between ileum and rectum. It was noted that the terminal foot of the ileum was thickened, indurated, and chronically inflamed. There were numerous peritoneal adhesions between loops of ileum. The mesentery of the affected ileum contained numerous large, oval, grayish nodes. A fistulous tract was present extending down along the right pelvic wall to the rectum about one inch above the anus.

Pathologic Examination, Gross: The resected intestine revealed chronic and acute

ulcerative terminal ileitis with multiple fistulae and subacute peritonitis. Eight centimeters from the ileocecal valve there was a perforation, about one centimeter in diameter, originating in the lumen of the ileum, and transversing the region of the mesentery. This was lined by reddish-gray granulation tissue. Four centimeters proximal to this fistula, and on the opposite side of the mesentery, there was another perforation of the bowel. The submucosa was thickened and showed numerous fistulous tracts. His postoperative course was complicated by a pneumonia which led to a lethal outcome after about two weeks.

Case 2.—Service of Dr. Richard Lewisohn. Hosp. No. 384578: H. B., male, age 21, had developed pain, redness and swelling in the perianal region two years previously. An abscess was incised, pus evacuated and the area gradually, but not completely, closed over. About 19 months later a probe was inserted into the persistent opening for a variable distance upward. The fistula refused to heal and continued to discharge thin purulent material.

About six months before admission (one and one-half years after the first appearance of the fistula-in-ano) he developed cramp-like supra-umbilical pains, nonradiating and unrelated to meals. This pain continued for five weeks with afternoon rises of temperature to 101° F. Appendicectomy was performed elsewhere and a "ruptured appendix with abscess" was found and drainage was instituted. The drain was removed in one week and the wound healed completely in 12 days. As he was about to be discharged, he developed pain in the right hip extending down to the thigh and associated with fever. The wound was reopened and again drained. A sinus to the abdominal wall formed which persisted for one month, but then closed; he was considered ready for discharge. At this time, he developed pain in the right lower quadrant where he noted a red, tender, painful, swollen area. This was incised and drained, and continued to drain up to the time he came under our observation, a period of four months. Six weeks before this period of observation the original incision reopened and remained as a draining sinus in the anterior abdominal wall. The tender mass in the right lower quadrant of the abdomen now reappeared.

Physical Examination.—The following significant features were noted: There were two incisional scars in the right lower quadrant. The medial scar was a three inch pararectus incision beginning just below the level of the umbilicus. It presented a pinhead-sized opening at its lower end. This orifice was surrounded by granulation tissue and exuded pus on pressure. The lateral incision paralleled Poupart's ligament and extended just above it two inches from the anterior superior iliac spine. It was surrounded by granulation tissue and showed a greenish, foul discharge. There was a mass just lateral to the pararectus incision over which tenderness and rigidity could be elicited.

On the left buttock, one inch from the anus and in the "4 o'clock position," was a crusted polypoid mass from which pus exuded. This was surrounded by an area of induration above which a second opening could be seen.

Injection of lipiodol into the upper abdominal sinus, with occlusion of the lower one, outlined several tortuous sinus tracts. The greater part of the lipiodol traveled downward and puddled in the pelvis.

Gastro-intestinal roentgenograms showed no evidence of a lesion of the stomach or duodenum. Observations of the small intestine, made at hourly intervals, showed the jejunal loops displaced to the left by a mass in the right iliac fossa. The terminal ileum was narrowed, irregular and markedly deformed, indicating the presence of a nonspecific ulcerating lesion involving the ileum and cecum.

Operation.—September, 1935: Dr. Leon Ginzburg. The ileum was cut across above the involved area, both ends were turned in and an ileotransverse colostomy was performed, thus excluding the mass from the fecal stream. His physical condition precluded any extensive exploration. His postoperative course was uneventful and the fistula-in-ano became asymptomatic. The discharge from the abdominal fistulae grad-

ually diminished, the fistula eventually closing. The fistula-in-ano closed spontaneously after the abdominal procedure, and has remained closed to date.

Case 3.—Service of Dr. Richard Lewisohn. Hosp. No. 407492: S. F., white, female, age 24, had been perfectly well until six and one-half years before coming under observation, when she suddenly developed diarrhea while undergoing reducing procedure. There were as many as 20 nonbloody movements daily, accompanied by severe perineal pain. Fever was absent. Six months after the onset, a "rectal cyst" was observed in another institution and incised. Four months after this operation the diarrhea subsided spontaneously. A report from the above hospital indicated that she had had a perirectal and a vulvovaginal abscess, both of which had been incised and drained.

Six months after the first operation, she developed an abscess of the right labia majora, which was incised and drained. Shortly thereafter a swelling of the right buttock developed, which was also incised and drained, as was also an abscess of the left labia. The surgeon reported these as simple ischiorectal abscesses. One and one-half years after the onset, and soon after the drainage of the second labial abscess, a rectovaginal fistula was found. This fistula was repeatedly operated upon, but never with successful issue. Feces were passed per vaginam and there was a persistent vaginal discharge. The failure of the rectovaginal fistula to close led to further investigation and it was found that there was a sinus tract in the rectovaginal septum which ran so far up that "it was necessary to leave the innermost portion of it behind because of the danger of entering the abdominal cavity." At this time a roentgenologic examination of the gastro-intestinal tract showed a stasis in the small intestine which was attributed to adhesions. Shortly thereafter, a celiotomy for "intestinal obstruction" was performed and an appendicectomy was carried out. No details of the intra-abdominal findings were given.

About ten months before coming under our observation, she was suddenly seized with severe abdominal pain and admitted to another institution. Reports from this hospital indicate that the patient apparently had had a partial intestinal obstruction, which was attributed to a "chronic adhesive peritonitis." During all the years of her illness, the patient had suffered from diffuse lower abdominal cramps which were present daily, and were accompanied by frequent recurrences of her watery diarrhea. In the five weeks preceding our observations, the cramps and diarrhea had increased to the point of necessitating hospitalization.

Physical Examination.—Mt. Sinai Hospital, New York: The patient was a well-developed and well-nourished young white female who presented a soft abdomen which was not distended or tender. There was a well healed midline hypogastric scar. No intra-abdominal masses were felt. There were several well healed scars in the perineum. Gynecologic examination showed a fistulous opening just within the vagina, about one-half inch above the fourchette, and leading into the rectum. Tuberculin tests were negative. Roentgenologic examination of the chest showed no abnormalities. Blood count showed only a mild secondary anemia. The blood Wassermann test was negative. Blood chemistry studies on admission were essentially normal. Examination of the stool showed the presence of occult blood but no ova or parasites. There were no serologic or cultural evidences of dysentery infection. Examination of gastric contents showed the presence of free acid. A gastro-intestinal series showed a dilatation of the distal jejunum and ileum, with evidence of an ulcerating lesion in the latter. Under observation, she progressively lost weight and had repeated episodes of fever and abdominal cramps.

Operation.—A typical regional ileitis, with marked enlargement of the mesenteric nodes and massive adhesions which matted together various loops of small intestine was demonstrated. A fistula was found connecting the ileum and sigmoid colon. This was divided, and the involved ileum, cecum and ascending colon resected and and ileotransverse colostomy performed. It was then found that there was a fistulous tract in the

mesentery extending down toward the pelvis. It was not explored further due to the duration of the operative procedure.

Following a prolonged postoperative convalescence, the patient was discharged much improved, but with the rectovaginal fistula still patent. It was considered wise to postpone repair of this fistula to a later time.

Discussion.—We may summarize these three cases as follows: In Case 1, a fistulous tract originated in the ileum and burrowed down into the pelvis, presenting itself as an abscess which reached to within an inch of the perineum before it was incised. The second case presented a small fecal fistula opening in the perineum. This was the result of an ischiorectal abscess which antedated the onset of the intestinal symptoms, but which eventually pointed to the presence of a regional ileitis. In addition, the patient presented two fistulae to the abdominal wall. The injection of lipiodol through the abdominal wall fistulae led to a cavity deep in the pelvis. A short-circuiting operation resulted in the spontaneous healing of all the fistulous tracts. In the third case, we were confronted with two fistulous tracts, both directed toward the pelvic floor—the one extending downward from the ileum to the ischiorectal fossa, the other extending upward from the perineum through the rectovaginal septum approaching the same area in the pararectal space.

In a recent analysis of the clinical histories of a series of 56 consecutive cases of regional ileitis, we were struck by the frequent occurrence of "fistula-in-ano" as narrated by the patient in discussing past gastro-intestinal disorders. Several of these patients presented perineal scars as evidence of previous surgical interference and in a few instances several nipple-like excrescences were found from which purulent material could be expressed. Objective data on such cases were observed in eight instances in this series, indicating an incidence of about 14 per cent.

Such perirectal or perianal suppuration would not be considered an unusual complication in a disease process so prone to manifest itself with diarrhea, were it not for its high incidence and unusual clinical features. In an analysis of 693 cases of chronic ulcerative colitis, Bargen found 26 cases of perirectal abscess and fistula-in-ano, which he considered to be due to infection of the crypts of Morgagni with subsequent invasion of the perirectal tissues and abscess formation. These figures would indicate an incidence of 3.7 per cent of fistula-in-ano or perirectal abscess in chronic ulcerative colitis. In a similar analysis of a mixed group of patients with various complaints referable to the anus, rectum or colon, he found fistula-in-ano to be present in 5 per cent of the total series.

The well-known tendency to intra-abdominal sinus and fistula formation shown by the pathologic process characteristic of regional ileitis, led us to believe that the perineal process might be another expression of the same burrowing capacity so frequently found in the production of fistulous connections between ileum and intra-abdominal viscera or abdominal wall.

These long or continuous fistulae are uncommon in comparison with the

more frequently observed simple, short fistula-in-ano. They are, however, exceedingly interesting and will bear careful clinical observation and recording. As a clinical manifestation, they frequently precede all symptoms which may cause suspicion of, or direct attention to the intestinal tract and particularly, the ileum.

For that matter, all fistulae which occur in association with ileitis may precede the consciousness of an intestinal disturbance. This applies to the fistulae in the abdominal wall which follow a futile exploratory operation, as well as to fistulae to segments of the colon, to the inguinal or lumbar regions, on fistula-in-ano. The original disease is of so subdued a nature and runs such a mild chronic course that the fistulae often precede the history or consciousness of diarrheal disturbances. In fact, of late we have seen ileitis with only constipation, but with fistulae-in-ano already recognizable.

Every case which presents itself with fistula-in-ano should deserve careful clinical preoperative study. The general conception that most fistulae-in-ano are tuberculous in origin is entirely erroneous; according to the pathologic records of the Mt. Sinai Hospital (Dr. Paul Klemperer), less than 1 per cent of granulation tissue removed at operation and subjected to study, reveals the tuberculous nature of such fistulous tracts.

Every case of fistula-in-ano is entitled to a proctoscopy, to rule out ulcerative colitis; to a careful roentgenologic study of the intestinal tract; to a roentgenogram of the chest to establish the existence or nonexistence of tuberculosis; and to a careful history and an even more careful physical examination.

In any case suspected of ileitis in which the roentgenologic findings are inconclusive and not convincing, the existence of one or more perianal fistulous tracts should materially support the diagnosis of an ileitis, providing of course that ulcerative proctitis or colitis is excluded.

Treatment.—Perianal fistulae, pararectal abscesses and rectovaginal fistulae originating from ileitis are best handled surgically by efforts directed to remove the primary disease. When the ileitis is entirely resected, the fistulae almost invariably close and heal permanently. Our experience with short-circuiting operations is insufficient, but we have reason to doubt that they will uniformly, or even in a majority of the cases, lead to healing of the fistulous tracts in the anal region; occasionally, however, such short-circuiting operations are successful in causing healing of the fistulae.

Persistent diarrhea is a deterrent to healing of such tracts; it is better to resect the ileitis in its continuity rather than leave a persistent focus of infection within the intestinal lumen with its recognized capabilities as a trouble-maker.

Where the fistulae fail to heal spontaneously after the ileitis has been properly handled, local excision of the tract, curettage and suture should be instituted, provided the bowel function has been restored to normal. In our experience the rectovaginal fistulae always require operation and cauterization, usually, unfortunately, with little success, so that many attempts are required before a satisfactory result can be obtained.