Spontaneous Resolution of Double Barrel Ileostomy and Complex Enteroatmospheric Fistula in A Single Patient; A Case Report

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Abbreviation:
ECF: Enterocutaneous Fistula; CECT: Contrast Enhanced Computed Tomography; BMI: Body Mass Index; EAF: Enteroatmospheric Fistula

1. Abstract
Ileostomy is a surgically created opening of the bowel onto the anterior abdominal wall to facilitate evacuation of bowel content in a controlled manner. All temporary ileostomy need surgical closure once the purpose is fulfilled.

We are presenting a case of a 24-year female who had spontaneous resolution of double barrel ileostomy with complex Enteroatmospheric Fistula. She had previously undergone exploratory laparotomy and double barrel ileostomy for ileal perforation peritonitis. Spontaneous resolution of ileostomy is a very rare event. Exact pathophysiology behind this event is unknown till date, although it is being attributed to those factors which favor the spontaneous closure of Enterocutaneous Fistula. This needs a considerable number of cases and work before coming to any conclusion.

2. Introduction
Intestinal stomas are surgically created opening of the bowel or urinary tract onto the anterior abdominal wall [1]. Ileostomy is an iatrogenic Enterocutaneous Fistula fashioned to facilitate evacuation of bowel contents in a controlled manner [2]. Once the purpose is fulfilled, temporary ileostomy needs closure [3]. There have not been many cases of spontaneous closure of ileostomy making this a very rare event [4]. Of all the complications of ileostomy, spontaneous resolution seems to be a boon to the patient, as it obviates the need for surgery. There is lack of evidence showing the etiology/pathophysiology favoring spontaneous closure of stoma [5]. Some literature has compared ileostomy as a controlled ECF. In some literature, pathophysiology contributing the spontaneous closure of ECF has been attributed to spontaneous closure of stoma [3, 4]. We are here presenting a case of a 24-year female who had spontaneous resolution of double barrel ileostomy and complex Enteroatmospheric Fistula.

3. Case Report
A 24-year gravida at 32 weeks’ period of gestation presented to our institute with a complaint of pain over lower abdomen for 5 days, which was acute in onset and gradually progressive. Later, the pain was diffuse all over the abdomen which was severe in intensity and aggravated on movements. There was a history of abdominal distension which was gradually progressive. It was associated with multiple episodes of vomiting which contained food particles. Patient was unable to pass stool for 4 days.

On examination, she had high grade fever and her other vitals were stable. Per abdomen examination revealed distended abdomen with tenderness all over the abdomen. Bowel sounds were present.
She delivered a live baby on the very next day of admission via spontaneous vaginal delivery. Till her 4th post-partum day, the distension and pain in the abdomen did not improve. There was persistent tenderness all over the abdomen. Chest x-ray revealed free gas under right hemi diaphragm which was initially absent at the time of admission. We, then, proceeded to exploratory laparotomy, where there was a gross peritoneal contamination along with thinned out ileal wall (~210 cm from duodenojejunal flexure) with multiple perforations along ~10 cm segment of its length. We resected that segment of the ileum and made a double barrel ileostomy. Histopathology showed focal transmural infarction of ileum. Further investigations to rule out inflammatory bowel disease or Tuberculosis was not done.

Hospital events

Day 2: Stoma was functioning; blackish discoloration of stoma was seen.
Day 5: Stoma was functioning; stoma wall was slightly retracted.
Day 8: Burst abdomen occurred, proximal segment was completely retracted but stool was coming out from the stoma site.
Burst abdomen was managed with Bagota bag. Patient was advised for high protein diet as there was hypoalbuminemia (albumin=2.1gm/dl).
Day 20: Patient had 2 ECFs in midline from which stool was coming out in addition to the stoma site (Figure 1).

We decided to discharge the patient after 2 months of hospital stay. She was called on a follow up after 2 months. At the time of follow up, stoma was completely retracted, but there was evidence of two low output ECFs on midline. She also gave a history of occasional passage of stool via anus (2 to 3 times) during that period. As there was no evidence of peritonitis on examination, we continued the same management protocol and postponed the surgery. She was advised to take high protein diet and was called on a further follow up after 2 months.

On the recent visit, midline was healthy, healthy scar was present at the stoma and drain site, and there was no evidence of the previous ECFs (Figure 2). She was taking a normal diet and passing stool and flatus via anus.

4. Discussion

Spontaneous resolution of ileostomy is a very rare occurrence. It has aroused much interest among surgeons as the exact mechanism behind it is unknown. In many literature, ileostomy is being compared to controlled enterocutaneous fistulas. We will here try to correlate the factors responsible for closure of Enterocutaneous Fistula with the spontaneous resolution of ileostomy.

The initial step of resolution is the retraction of stoma as evident by Salvadena in his literature. He had shown that the incidence of retraction decreases with time, which was 8% by day 10, 7% by 3 month and 1% by 1 year. Similarly, the time of retraction was early in our case [6].

There are various etiologies of retraction of stoma but despite the lack of pathophysiology supporting the spontaneous resolution of stoma, it has been mostly attributed to factors responsible for closure of controlled ECFs.

Among the stoma complications, retraction may be considered advantageous, which occurs in 0 to 40% of cases. The cause for retraction may be the result of too much tension on bowel or its mesentery. This tension may be due to tethering of the mesentery

Figure 1: Retracted proximal part of stoma with multiple enterocutaneous fistula

Patient was managed on the line of ECF.

At 1 month: It was observed that the amount of stool from the stoma site gradually decreased while that from midline increased considerably.

At one and a half month: ECF’s on the midline were having controlled output, stool coming out from the stoma site decreased to only about 10-50 ml/day.

Figure 2: Healed stoma site, midline and drain site
or insufficient mobilization of bowel or sites and the types of stomas. Incidence of retraction was found more among loop stomas as compared to end stomas [7]. Other factors such as high Body Mass Index (BMI), steroid use, malnutrition, diabetes and smoking have also been implicated for the retraction of stomas [5].

Enterocutaneous Fistulas are most commonly iatrogenic, usually the result of a surgical mishap. On reviewing the literature, it becomes clear that healing of an EC fistula is favored by the absence of foreign body, radiation, infection or inflammation, epithelialization, neoplasm and distal obstruction [4, 8]. Similarly, those Enterocutaneous Fistulas with low output (<200mL/24h), <50% of the involvement of bowel circumference and fistulous tract more than 2.5 cm are presumed to be healed spontaneously.

In our case, our patient also had a complex Enteroatmospheric Fistula, which can be broadly defined as a fistula in an open abdomen or occurring in the wound of a dehiscent laparotomy scar.

This type of fistula is also associated with relatively lesser probability of spontaneous healing. According to a report by Visschers et al., spontaneous closure of these type of fistulas is five times less likely which was the just the opposite finding in our case and thus can be considered as one of the rare events [4].

Keeping all these factors in mind, an ileostomy can be considered as a post-operative, low volume, simple and superficial type of Enterocutaneous Fistula and can expect all ileostomies to regress spontaneously without any surgical intervention theoretically.

The spontaneous closure of a stoma can be considered as the final outcome of a gradually progressive stomal retraction. In our case, retraction of stoma was not associated with peritonitis and patient was passing stools and flatus per anum. Therefore, an approach of watchful waiting was adopted and revision surgery was postponed. However, factors pertaining to this particular case which led to spontaneous closure of stoma remain undiscovered. The possibility of entero-enteric or entero-colic fistula bypassing ileostomy leading to spontaneous closure was made in some reported cases but needs additional investigations like CECT abdomen or other contrast studies to support this [4].

Approximately 30% of patients develop incisional hernia requiring repair after surgical stoma closure. Owing to this fact, this incidence may even increase in case of spontaneous closure of stoma, where the defect of the abdominal fascia is open and thus may need regular follow up [5].

5. Conclusion

Owing to its rare occurrence, exact pathophysiology of spontaneous closure of ileostomy is still inconclusive and debatable. This needs a considerable number of cases and work before coming to any conclusion. Whether we may want to call it a rare ‘complication’ or a ‘boon’, ultimately patient is the real beneficiary.

References