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**Ileo-anal pouch excision: A review of indications and outcomes**

Byrne CM *et al.* Review of Ileo-anal pouch excision

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**Abstract**

Restorative proctocolectomy (RP) is the surgical treatment of choice for ulcerative colitis (UC) and patients with familial adenomatous polyposis (FAP). A devastating complication for both patient and surgeon is failure of the pouch that requires excision. There is currently no single paper in the literature that consolidates the indications for ileo-anal pouch excision and the subsequent outcomes following this procedure. A literature search was carried out to identify articles on RP and ileal pouch-anal anastomosis. The main search terms used were “RP”; “ileal pouch-anal anastomosis” or “ileal reservoir” or “ileal pouch”; “failure of ileal pouch-anal anastomosis” and “excision of ileal pouch-anal anastomosis”. The search was completed using electronic databases MEDLINE, PubMed and EMBASE from 1975 to June 2014. Characteristics of patients with pouch failure differ between institutions. Reported overall excision rates of the pouches vary and in this review ranged from 0.93% to 12. 8%. Age and lower institutional volume (less than 3.3 cases) were independent predictors of pouch failure; however surgeon case load was not. The main reasons identified for excision are sepsis (early cause), Crohn’s disease and poor functional outcomes (both late causes). Pouch cancers in UC and FAP are still rare but 135 cases exist in the literature. The most common complication following excision is persistent perineal sinus. The decision to excise a pouch should not be taken lightly and an awareness of the technical pitfalls and complications that can occur should be fully appreciated.

**Key words:** Ileo-anal pouch; Pouch failure; Pouch excision; Indications and outcomes

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**Core tip:** There is currently no single paper in the literature that consolidates the indications for ileo-anal pouch excision and the subsequent outcomes.Reported overall excision rates vary and in this review ranged from 0.93% to 12. 8%. Age and lower institutional volume (< 3.3 cases) were independent predictors of pouch failure; however surgeon case load was not.Main reasons identified for excision are sepsis (early), Crohn’s disease and poor functional outcomes (both late causes). Pouch cancers in ulcerative colitis and familial adenomatous polyposis are rare but 135 cases exist in the literature. An awareness of the technical pitfalls and complications that can occur should be fully appreciated.

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**INTRODUCTION**

Restorative proctocolectomy (RP) is the surgical treatment of choice for ulcerative colitis (UC) and selected patients with familial adenomatous polyposis (FAP). The RP procedure can significantly improve a patient’s quality of life[1] but it is also associated with considerable postoperative morbidity, with up to 50% of patients reporting complications[2]. However, 30 day and 1 year mortality remain low at 0.5% and 1.5% respectively[3].

A devastating complication for both patient and surgeon is failure of the pouch that requires either excision or permanent diversion with a loop ileostomy or permanent Brooke ileostomy. Characteristics of patients with pouch failure vary between institutions. Published pouch excision (PE) rates after RP range from 2% to 22%[4].

The aim of this review was to identify the reasons for RP failure that resulted in excision and to discuss the difficulties associated with removal.

**RESEARCH**

A literature search was carried out to identify articles on RP and ileal pouch-anal anastomosis. The search was completed using electronic databases MEDLINE, PubMed and EMBASE from 1975 to June 2014. The main search terms used were “RP”; “ileal pouch-anal anastomosis” or “ileal reservoir” or “ileal pouch”; “failure of ileal pouch-anal anastomosis” and “excision of ileal pouch-anal anastomosis”. Series included patients with UC and FAP and concentrated on RP related complications and the technical aspects of excision. Additionally, references and citations from all retrieved articles were analysed for identification of similar reports. Exclusion criteria included papers published in languages other than English. Two reviewers independently screened studies for inclusion and when duplicated or updated cohorts were identified, only the most recent study was included.

**DISCUSSION**

***Pouch failure***

Failure of RP is defined as permanent diversion or PE.

The reasons for failure and subsequent excision can be broadly categorised into the following technical or disease-related causes: (1) Sepsis (including anastomotic leak, fistula, pouch-vaginal fistula); (2) Mechanical or functional problems (stricture, outlet obstruction, incontinence); and (3) Disease-related failure [Crohn’s disease (CD) being the most common[5]).

Early onset pouch failure, *i.e.*, failure within 12 mo is typically caused by surgery-associated complications such as pelvic sepsis[6], anastomotic stricture and separation[7] and pouch sinus and fistula[8]. Reasons for late onset pouch failure (after 12 mo) include CD of the pouch[7,9-11], chronic pouchitis[12], refractory cuffitis, pouch strictures, prolapsed pouch, refractory pouch-vaginal fistula and carcinoma.

Further documented risk factors are mucosectomy[13,14], anal pathology, abnormal anal manometry before surgery[7], and experience of colorectal surgeons in performing pouch surgery[7,15]. Large institutional case loads are associated with reduced reoperation and failure rates[16].A study including 5771 patients from 154 English National Health Service Trusts also found that there were significant relationships between 365-d mortality and failure rates with case volume. Age and lower institutional volume (less than 3.3 cases annually)were independent predictors of pouch failure; however individual surgeon case load was not[3]. Divergence in failure rates that occurs between high (more than 8.4 cases annually) and low volume institutions appear to occur beyond the peri-operative period. Low volume centres operated on more patients with CD; therefore, poor preoperative histological diagnosis and case selection may contribute to the higher failure rates rather than operative technique or surgical skill and technique[3].

***Reasons for PE***

**Sepsis:** Many colorectal surgeons will routinely elect to divert an RP with a loop ileostomy given the devastating complications that can occur after one-stage RP. The omission of an ileostomy still remains controversial[17] as published clinical trials comparing RP with and without a covering ileostomy lack the statistical power to provide a definitive recommendation[18]. However, a review of 17 studies comprising 1486 patients reported that the rate of pelvic sepsis was in fact significantly lower in patients with a temporary ileostomy[19]. In this review of PEs we found that most institutions opted for proximal diversion for their primary restorative RP procedures[7,20-30].

Pelvic sepsis rates are reported to be 5% to 19% of cases[6,24,25]. A meta-analysis by de Zeeuw *et al*[31] which included 14,996 patients found a pooled incidence of pelvic sepsis of 7.5% (95%CI: 6.1-9.1). Anastomotic leak occurs in 7% to 15%[32,33] and both pelvic sepsis and leaks can subsequently result in a re-operation rate of 24% to 63% in this cohort of patients[20,32-34]. The management of pelvic sepsis is multi factorial which includes clinical stability of the patient, the extent of sepsis (localised or generalised, abscess, collection, and peritonitis) and services available locally. Management can involve conservative treatment with antibiotics and radiological/trans-anal drainage depending on the position of the collection; or it can also be treated surgically with salvage operations or PE as a last resort. The St. Mark’s Hospital[4] series included 996 patients who had undergone primary RP and included a further 245 patients who were referred for salvage procedures over a period of 25 years (1977-2002). Pelvic/perineal sepsis accounted for 51% (35/68)[4,35] of PEs in their series. Ninety-seven percent of patients who had their primary RP at St. Mark’s had at least one or more salvage procedures before excision occurred (median 2; range 0-11). Overall 22.4% of their pouches excised were done so within 12 mo with the remaining excised at a median of 50 mo (range, 13-230 mo)[4].

The Cleveland Clinic series (1985-2009) reported a similar outcome with sepsis/fistulae contributing to 40% of their overall PEs (110/1965 patients)[26]. The Mayo Clinic (1981-1994) had a pelvic abscess rate of 4.9% (73 patients) with 19% (14/73) of these patients subsequently undergoing PE [21]. Pelvic sepsis accounted for 45% of all pouch failures within 2 years at the Mayo Clinic but for less than 2% of all subsequent failures (over 2 years)[36]. MacRae *et al*[6] (Toronto 1981-1992) overall pouch failure rate was 10.5% (58/551); 84.5% of pouches that failed required excision. Leak from the pouch or ileo-anal anastomosis were found to be significantly associated with PE *P* < 0.0001 and *P* < 0.001 respectively, as was no proximal diversion with an ileostomy *(P* < 0.01)[6]. Identification of pouch failure following pelvic sepsis is largely dependent on the duration of follow up. A meta-analysis from the Netherlands including 43 studies reported that pooled incidence rates increased from 6.8% to 8.5% if patients were followed up for at least 5 years. There was also no major difference in failure rates secondary to sepsis between series that included 200 patients and those that included 1200 patients[24].

**CD/ Fistula:** Permanent pouch failure occurs significantly more often in patients with CD when compared to those with UC (36.8% *vs* 1.4%)[37]. Reported post-operative diagnosis of CD in the current literature ranged from 0.7% to 4%[4,6,24,29,38,39] with 43% of all PEs at the Cleveland clinic found to have a final histological diagnosis of CD[26]. A diagnosis of CD can be associated with a 4-fold increase in the likelihood of pouch failure[7] and failure rate for inadvertent RP for CD can be as high as 50%[12,27,29,39]. A pre-operative diagnosis of CD is considered a contra-indication to RP as it is believed that disease recurrence and potential for fistula is high and that ultimately PE may be necessary[40]. This is certainly reflected in the literature where it has been reported that the principal reason for pouch failure in CD patients is persistent sepsis secondary to fistulating disease. In a series published from the Lahey Clinic, 40% of their pouch failures were due to fistulating CD[38] and unsurprisingly 90.9% of patients from the Mayo Clinic with CD and whom developed complex fistulating disease (median 29 mo; range, 3-60 mo) post RP required excision[29].The Cleveland study reflected the experience of other centres and multivariate analysis found perineal (adjusted HR 3.198, 95%CI: 1.986-5.148, *P* < 0.001) and vaginal fistulae (adjusted HR 7.491, 95%CI: 3.031-18.514, *P* < 0.001) to be significant predictive factors of pouch failure[7]. No single risk factor for failure on multivariate analysis was statistically associated with either the early-onset (less than 12 mo) or late onset fistula (over 12 mo), and no difference in pouch failure was found between either two groups[41].

**Poor functional outcome:** Symptomatic anastomotic stricture is associated with an increased likelihood of pouch failure *(*adjusted HR 2.692, 95%CI: 1.824-3.971, *P* < 0.001)[7] and rates of excision due to anastomotic stricture ranged from 2.03% to 27.3%[4,6,12,38,39]. Poor function is often referred to as “outlet obstruction or incontinence.” At the Cleveland Clinic 30% of pouches were excised due to stricture or poor function[7] with a similar excision rate for poor function of 35% observed at St. Mark’s Hospital[4]. In the series by Prudhomme *et al*[27] 50% of pouches excised was due to poor function but typically each patient who had undergone excision had more than one complication and therefore poor function is likely multi-factorial.

**Pouch ischaemia:** Pouch ischaemia as a result of technical failure will usually present early and is now regarded as an avoidable complication. Korsgen *et al*[12] reported a relatively high excision rate due to ischaemia (26.1%, *n* = 6/23) however only two of these cases were due to technical problems (extensive vascular mobilisation, small bowel haematoma). They reported two late cases due to rotation injury along the long axis of the small bowel mesentery and ileoanal anastomosis[12].Karoui *et al*[4] had an excision rate due to pouch ischaemia of 1.5% and Farouk *et al*[21] a rate of 14.3%.

**Chronic pouchitis:** Pouchitis is rarely seen in those patients with FAP and is more common in UC patients[42]. The incidence of pouchitis increases from 40% of patients having one episode in first 10 years to 70% within 20 years[18]. The pooled incidence of at least one episode of pouchitis was 18.8% (95%CI: 15.7-22.4) from a meta-analysis of 43 studies[24]. However, current evidence suggests that pouchitis, as a single entity, is rarely the reason for pouch failure[4,6,8,30,32,34,38,43]. Approximately 5%-10% of patients develop chronic pouchitis that requires long term therapy and a small minority will have pouchitis that is refractory to medical treatment. This is the subset of patients that should be referred to a Colorectal Surgeon for a discussion surrounding permanent diversion or excision[44]. The rates of excision of a pouch for pouchitis alone were between 7.4% and 22.9%[4,6,8,20,22,26,30,35]. However, one Canadian study reported an excision rate of 54.5% (6/11) due to intractable pouchitis in their UC cohort who had undergone excision[45].However this was in an era where the use of long term anti-biotics and biologicals were still under evaluation.

***Dysplasia/cancer***

Both UC and FAP predispose to neoplasia within the pouch. It is useful to consider these diseases separately.

**UC and cancer:** A recent study by Wu *et al*[46] (3203 patients) reported the cumulative incidence for pouch neoplasia at 5, 10, 15, 20 and 25 years after pouch construction in UC patients were 0.9%, 1.3%, 1.9%, 4.2% and 5.1% respectively. They also concluded that those patients with a final diagnosis of pouch adenocarcinoma when compared to those with dysplasia tended to be older (*P* = 0.04) and had a longer duration of diagnosis of IBD or pouch construction prior to the detection of neoplasia (*P* = 0.007 and *P* = 0.0013). Eleven out of fourteen patients with adenocarcinoma had resection and PE with curative intent (APR with end ileostomy *n* = 8, APR with Kock pouch *n* = 2, palliative resection with end ileostomy *n* = 1) and 2/12 with high grade dysplasia had excision (APR with end ileostomy *n* = 1 and APR with Kock reservoir *n* = 1).The prognosis for pouch adenocarcinoma is poor and the anal transition zone was the most common site in the Cleveland Clinic Series[46]. The occurrence of neoplasia in patients with RP is not eliminated by mucosectomy[19,47].

However it is not clear whether retaining the anal mucosa using the double stapling technique and allowing the mucosa to be sampled is superior or inferior to mucosectomy. This was reviewed by M’Koma *et al*[48] in 2011, who found 43 cases of pouch cancer related to UC. Thirty two had transition zone carcinoma, 28 of whom had had a mucosectomy. There were also 11 cancers within the body of the pouch body[48]. Patients with primary sclerosing cholangitis and inflammatory bowel disease are at an increased risk of colorectal neoplasia. Imam *et al*[49] found a cumulative 5-year incidence of pouch neoplasia of 5.6% (95%CI: 1.8%-16.1%)[49].However, there is still insufficient evidence to implicate PSC as a risk factor for the development of dysplasia and carcinoma of the pouch[49].

**FAP and cancer:** Incidence of adenomas (within pouch) in patients with FAP varied from 6.7% to 73.9%, age of the pouch is an important risk factor: 7%-16% at 5 years, 35%-42% after 10 years and 75% after 15 years. However, only 23 cases of ileal pouch carcinoma have been recorded in the literature to date[50]. Thus data suggests that the body of the pouch needs to be reviewed after 5 years in patients with FAP. In the review by Smith *et al*[50] in 2013 there were 92 cancers in total of which 23 occurred within the body of the pouch and or in the anal canal mucosa or cuff again, suggesting lifetime surveillance for FAP pouches. In the St. Marks series two pouches were excised as a result of unrecognised cancer in the rectum at the time of original RP construction (2.9%)[4], and 6.1% of pouches at the Cleveland Clinic were excised due to neoplasia of the pouch or rectal cuff[26]. Prudhomme *et al*[27] reported PE as a result of desmoid tumours in a total of three cases[27]. Other reasons for PE are outlined in Table 1.

***PE***

Excision of a failing pouch sacrifices a significant length of terminal ileum and there are a small number of papers that suggested transformation of a pouch to continent ileostomy (Kock reservior) may be a suitable alternative[51,52]. This would not be the authors operation of choice. However, current indications include patients who require a panproctocolectomy but cannot have a pouch constructed, those patients with failed RP who are not candidates for redo surgery and those with a Brooke ileostomy that is adversely affecting the patients quality of life[53].Ecker *et al*[52] successfully converted 4 ileo-anal pouches to a kock reservoir and the indication was functional disturbance that could not be corrected surgically. Hultén *et al*[51] had a series of 5 patients who had transformation of their pouch for pouch-vaginal fistula considered unsuitable for local revision, unsatisfactory function and unacceptably high defecation frequency. Performing a permanent ileostomy above a pouch left in-situ is another reasonable alternative when PE is not feasible or recommended particularly as this is not associated with neoplasia[54]. Once the difficult decision has been taken to excise a pouch, a comprehensive and carefully constructed management plan/surgical strategy must be implemented. The urgency with which the operation is performed will depend on the indication for excision and the clinical stability of the patient. If time permits, the authors feel that these patients should be discussed in a multi-disciplinary environment with further discussions with the patient in an appropriate setting, ideally with their own network of support available. Patients often give weight to other values besides physical health and the disappointment after unsuccessful restoration of intestinal continuity and the prospect of excision can be devastating. Lepisto *et al*[8] reported that quality of life scores are lower in PE groups when compared to the general population and this is often due to physical impairment and social restrictions[8].Transparency is of paramount importance in order to meet patients’ expectations and the appropriate support network should be in place before excision takes place. The network should include aspects of metabolic, nutritional and psychological support as short gut may be a problem in patients some of whom will be stoma averse.

**THE OPERATION**

There is a paucity of information in the literature about the operative strategies for PE but the approach taken for re-operative/salvage surgery should be adopted for excision surgery (preservation of bowel length is a principle as is nerve/ureteric preservation). An exhaustive evaluation with imaging such as computed tomography, magnetic resonance imaging and endoscopy are required. When salvage surgery is undertaken at the Cleveland Clinic patients are placed in the Lloyd-Davies position and both the abdomen and perineum are prepared and draped. The abdomen is entered via the previous incision; the pouch mobilised to the levator ani muscles using sharp dissection and the pouch is subsequently excised. In addition, intra-operative urethral stents[55] are often necessary and would certainly be advocated by many colorectal surgeons with experience of the difficulties encountered in the re-operative pelvis. PE may be required with a temporary proximal diversion in place. This may have been for a salvage procedure or to manage sepsis, and once all sepsis has resolved (3-6 mo), further surgery can be considered[5]. Fifty-one percent of patients in the St Mark’s series had a diverting stoma for sepsis, pouchitis and poor function prior to excision[35]. PE and perineal closure can be performed using an extrasphincteric (ES), intersphincteric (IS), or sphincter preserving (SP)[26].Nisar *et al*[26] from the Cleveland Clinic prefer IS dissection and reserve ES closure for cancer or extensive perineal sepsis and authors from this institution adopt this same principle. SP closure is employed when restoration of intestinal continuity may be a future option[26]. Prudhomme *et al*[27] performed 24 PEs with sphincteric dissection as follows: total sphincteric 10/24 (levator ani muscles were closed and the subcutaneous and skin layers were closed), intersphincteric 10/24, no sphincteric dissection 4/24[27]. The St. Mark’s operative strategy for PE is a combined abdominoanal approach with dissection maintained close to the pouch in order to minimise risk of damaging pelvic nerves. Dissection is commenced posteriorly after entering the presacral space behind the small bowel mesentery and continued caudally, laterally and then anteriorly to the level of the pelvic floor. The ileoanal anastomosis is disconnected and along with the anal canal is removed via an intersphincteric dissection. If there is evidence of pelvic sepsis the practice was to curette any granulation tissue and the perineal wound was left open in all cases. Ninety percent of these patients had a Brooke ileostomy with the remaining 10% (7/68) having a continent Kock reservoir[4]. Prudhomme *et al*[27] opted also for Brooke ileostomy in just over 90% of their patients and performed a Kock reservoir in only two patients[27].

There is a lack of studies that report the outcomes after excision of a pouch and this area needs more study[26]. Complications can be classified as early (within 30 d of surgery)or late. In the St Mark’s series[4] 25% of patients (*n* = 17) had immediate post-operative complications; one death. Seventeen patients had one or a combination of the following: sepsis (peritonitis, abdominal wound, pelvic abscess, peristomal abscess), bleeding, intestinal obstruction deep vein thrombosis. Nineteen percent overall required further surgery. Median duration of follow-up after excision was 79 (range, 3-312) mo and during this period 53.7% (95%CI: 41-66%) were readmitted for late complications. The risk of readmission from the time of the PE was 38% (95%CI: 27-51%) and 58% (95%CI: 45-72%) at 1 and 5 years respectively. Perineal complications such as persistent perineal sinus (PPS) are the most common late complications. Complete healing can eventually be achieved in the majority of cases. However up to 10% of patients can still have PPS at 12 mo[27,35]. The Mayo Series had a 40% rate of PPS six months post-operatively and all of these patients required at least one further procedure with two requiring a laparotomy. Interestingly no correlation was found between indication for excision and PPS[4]. Despite an identical rate of PPS in the St Mark’s series, they stated that “PE for fistula or abscess in the pelvis or perineum was an independent predictor for PPS”[35]. The Cleveland Clinic had the largest series of PEs (110 patients, 48% CD) and had a PPS rate of 39.8% with an overall healing rate in this cohort of 80.7% with further procedures. They also found no association with PPS and closure technique or eventual healing *(P* = 0.37 and *P* = 0.94 respectively*)* and no significant difference in PPS was found between CD and non-CD patients. In the CD cohort where sphincter preservation was attempted four patients developed PPS. These patients achieved complete healing when sphincters were excised[26].This outcome was also reflected in the series by Prudhomme *et al*[27] who reported that the highest rate of PPS was in CD patients and complete healing was achieved when total sphincteric excision was subsequently performed[27].A small but significant difference was observed in quality of life scores (SF-12 Questionnaire) for both mental (PPS 43.5 ± 10.9 *vs* No PPS 50.9 ± 9.1, *P* = 0.038) and physical (PPS 40.8 ± 10.5 *vs* No PPS 48.0 ± 8.9, *P* = 0.037) components at latest follow up when patients with PPS were compared to those without[26].

Further uncommon, late complications included small bowel obstruction, ileostomy retraction, incisonal hernia, enterocutaneous fistula from Kock reservoir and short bowel syndrome*.* Incidence rate of persistent impotence was 7.14% (2/28 males) which is higher than rates described following an initial RP[4].

***Overview***

Pouch surgery has been ongoing for over 35 years since the first RP in the 1970’s[56] and the authors predict an epidemic of problematic pouches which may be attributable to the duration that pouches have remained in situ. Failure of an RP that then requires PE is a devastating complication and subsequent management of a failed pouch should be entirely patient focused. Pooled rate of pouch failure is 4.3% (95%CI: 3.5-6.3)and when compared to studies before the year 2000, a reduction of 2.5% is observed in the pouch failure rate *(P* = 0.0038)[31]*.* Reported overall excision rates of the pouches vary and in this review ranged from 0.93% to 12.8%[3,4,6-9,12,20,21,38,57]. The main reasons identified for excision are CD and poor functional outcomes (both late causes). Pouch cancers in UC and FAP are still rare but now number 135 in the literature[48,50].

The complexity of PE and the meticulous strategy and resources that are required strengthens the current discussion for centralisation of such services in order to deliver the best service possible to patients. We believe that pouch surgery should be performed by Colorectal Surgeons with a specialised interest in IBD Pouch Surgery with adequate local resources to appropriately manage and support these patients. The decision to excise a pouch should not be taken lightly and an awareness of the technical pitfalls and complications that can occur should be fully appreciated.

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**Table 1 Other reasons for excision**

|  |  |  |
| --- | --- | --- |
| **Ref.** | **Total number of excisions** | **Reasons for excision**  **(% of total excision)** |
| Farouk *et al*[21], 1998  Mayo Clinic  *n =* 1508 | 14 | Pouch ischaemia (14.3%) |
| Karoui *et al*[4], 2004  Cleveland Clinic  *n =* 1241 | 68 | Pouch ischaemia (1.5%)  Intra-abdominal bleeding (2.9%)  Redo operation and insufficient length on mesentery (1.5%) |
| Körsgen *et al*[12], 1997  Birmingham  *n =* 180 | 23 | Pouch ischaemia (26.1%) |
| Lepistö *et al*[8], 2002  Helsinki  *n =* 486 | 24 | Adrenal insufficiency and dehydration (4.2%)  Patients fear of incontinence after stomal closure (4.2%)  Perianal pain (4.2%) |
| Wexner *et al*[30], 1990  Minneapolis  *n =* 180 | 14 | Severe post-operative haemorrhage (7.1%) |