

# Temporal Trends in Ileoanal Pouch Surgery for Paediatric onset Ulcerative Colitis in England from 1997 to 2015 using Hospital Episode Statistics

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**Funding:** The views expressed are those of the author(s) and not necessarily those of the NHS, the NIHR or the Department of Health. George Bethell is funded by the National Institute of Health Research Academic Clinical Fellow programme. JJA is funded by an ESPEN personal fellowship and by an ESPR post-doctoral grant.

## **Abstract**

### **Introduction**

Ileal pouch-anal anastomosis (IPAA) following colectomy for ulcerative colitis (UC) achieves restoration of intestinal continuity with potential return of continence. It is undertaken relatively infrequently in children. We aimed to investigate the national frequency of IPAA in paediatric UC and report outcomes useful for surgeon/centre benchmarking.

### **Methods**

Hospital Episode Statistics data were obtained for all admissions in England (1997-2015) in children (<18 years) who underwent IPAA for UC using OPCS-4 procedural codes. Surgeon specialty, readmission and reoperation rates were identified. Data are median (interquartile range).

### **Results**

UC was diagnosed in 7604 children in whom 346 (4.6%) underwent IPAA at age 15 (13-17) years. Laparoscopy was used in 55 (15.9%) cases and in the most recent 10 years more commonly by specialist paediatric surgeons (SPS) than general surgeons (GS) (34.3% vs 14.7%,  $p=0.001$ ).

National frequency of IPAA ranged from 12-34 annually. Where speciality was available, 95/342 (57%) cases were undertaken by GS and 147/342 (43%) cases by SPS. The proportion of cases undertaken by SPS increased significantly compared to GS over the study period,  $p=0.0003$ .

Post-operative length of stay was 8 (6-11) days. During the index admission, unplanned return to theatre was required in 25/346 (7.2%). Following discharge 58 (16.8%) were readmitted within 30 days. Overall return to theatre rate within 30 days of pouch surgery was 11.0% (38/346).

### **Conclusion**

IPAA for UC within childhood is undertaken infrequently in the England, with a shift towards SPS undertaking surgery. These data can be used by surgeons to benchmark outcomes.

**Keywords:** Ulcerative colitis, Ileal Pouch-Anal Anastomosis, Colectomy, Outcomes.

**Level of evidence:** IV

### **Highlights:**

#### **What is currently known about this topic?**

- About 10% of children with ulcerative colitis undergo colectomy before their 18<sup>th</sup> birthday. Ileal pouch-anal anastomosis (IPAA) allows restoration of intestinal continuity but is undertaken infrequently in children.

**What new information is contained in this article?**

- This population based study found the median length of stay following IPAA was 8 days, with readmission required in 16.8% and return to theatre in 11%. These outcomes can be used for surgeon/centre benchmarking.

## 1. Introduction

The incidence of paediatric inflammatory bowel disease (PIBD) is increasing with the phenotype remaining equally severe (1)(2). In paediatric ulcerative colitis (UC), most patients present with extensive inflammation, and compared to adult-onset UC, the disease course has increased severity (3,4). An estimated 10% of paediatric UC patients will undergo subtotal colectomy during childhood, with many more requiring surgery following transition to adult services (5).

Patients with UC may undergo colectomy for either acute severe colitis, or more commonly as a planned procedure for treatment-refractory disease (6). In both cases there are frequently post-operative problems, with early complications faced in 12-75% of patients and late complications in 19-73% of cases (5,7,8). In addition, re-operation rates are high, occurring in up to 54% of children (8). These factors, coupled with the need for holistic management of growth, pubertal development and education, make timing of surgery of key importance in these children and young adults. Typically, European centres will opt for 2 or 3 stage procedures, with separation of ileal pouch-anal anastomosis (IPAA) from the colectomy (5). This allows for a period of stability following surgery where nutritional status can be optimised and education prioritised (9,10). In contrast centres from the United States can be more likely to conduct single stage procedures (11).

The rates of pouch surgery in children is highly variable, a recent British survey identified a huge variability in the number of IPAA procedures by centre, alongside the indication for surgery (12). Whilst it is clear that specialist paediatric surgeons (SPS) are performing IPAA procedures, it remains uncertain as to the outcomes related to these surgeries, with timing and surgical personnel being variable between centres. Additional uncertainties in practice include the involvement of adult general surgeons (GS), the absolute number of procedures performed, and the rates of complications following surgery at population level.

Outcomes for pouch surgery remain variable and up to 15-30% of pouches will fail in the long term. There are important impacts on women's fertility, with a relative risk of infertility of 3.9 in recent

meta-analysis suggesting the importance of IPAA formation after completing a family where feasible, and faecal incontinence, with rates are up to 25% (13,14).

In this study we aimed to utilise Hospital Episode Statistic (HES) data to detail temporal trends in paediatric IPAA surgery in England, observe short-term complication rates and understand the role of SPS and GS in performing these procedures.

## **2. Methods**

### **2.1 Data collection**

Hospital Episode Statistics (HES) data were obtained from NHS Digital for an 18 year period from April 1997 to April 2015, for children aged less than 18 years old. This included all hospital admissions during that timeframe for any child with a diagnosis of inflammatory bowel disease - identified with ICD-10 coding. UC was identified using the ICD-10 code K51.

IPAA formation was identified using the Office of Population Censuses and Surveys Classification of Surgical Operations and Procedures, 4th revision, (OPCS-4) codes G72.5, H04.2 or H29.1-4. Colectomy was identified using the OPCS-4 codes H04, H05, H11 and H29. Laparoscopic procedures were identified with the OPCS-4 code Y75. OPCS-4 codes used to identify intervention due to complications are shown in the annex.

Data analysis and linkage was undertaken by the first author. Duplicated episodes were identified using HESid, admission date and discharge date to ensure admissions, and the procedures that occurred during them, were only counted once.

### **2.2 Outcomes**

Outcomes of interest were - length of stay after pouch surgery, unplanned invasive intervention within initial admission, re-admission within 30 days of IPAA and unplanned invasive intervention on re-admission.

## **2.3 Ethical approval**

Approval to access and use HES data for this study was obtained during the application process to NHS digital. Additional ethical approval was not required as this study uses existing data which is fully anonymised.

## **2.4 Statistical analysis**

Statistical analysis took place using StataSE v15 (StataCorp LLC, Texas, USA). Chi-squared was used for categorical data 2x2 analysis and Mann Whitney U was used for non-parametric continuous data. Chi-squared for trend was used to compare number of cases undertaken over time. Data are reported as median with interquartile range, or number with percentage as appropriate.  $P < 0.05$  was considered significant.

## **3 Results**

### **3.1 Cases identified and surgery undertaken**

Over the 18 year study period, UC was diagnosed in 7604 children and 782 (10%) underwent subtotal or total colectomy (with or without IPAA) at a median age of 15 (IQR 13-16) years. There were 346 (4.6%) IPAA's occurring at a median age of 15 (13-17) years. Laparoscopy was used in 55 (15.9%) cases at time of IPAA surgery. In the most recent 10 years of data (2006-2015), laparoscopic IPAA was more commonly performed by SPS than GS (34.3% vs 14.7%,  $p = 0.001$ ). The number of IPAA's performed across the UK ranged from 12-34 per year over the time period studied.

### **3.2 Surgeon specialty**

Where speciality was available ( $n = 342$ ), 195 (57%) cases were undertaken by GS and 147 (43%) cases were undertaken by SPS. Cases undertaken by a general surgeon were older than those performed by a paediatric surgeon (16[15-17] vs 13[11-15] years,  $p < 0.001$ ) and cases undertaken by SPS increased significantly compared to GS over the study period,  $p = 0.0003$  (figure 1).

### **3.3 Timing of colectomy**

To explore patient journey, children born prior to the start of the dataset were excluded for this section of the analysis, leaving 76 who had undergone pouch surgery. This was to ensure that colostomy formation had not occurred prior to the start of the dataset which would misclassify these children. Of these, 35 (46%) children underwent colectomy followed by pouch formation (three stage procedure) at a median of 326 (42-5239) days later and 41 (54%) underwent simultaneous colectomy and pouch surgery (two stage procedure).

### **3.4 Post-operative course and complications**

Post-operative length of stay was 8 (6-11) days and the median length of stay calculated per year decreased overtime, figure 2. During initial admission, there were 25 (7.2%) children with an unplanned subsequent invasive procedure (table 1) and 58 (16.8%) children were re-admitted within 30 days of IPAA. Age of child was similar between those re-admitted or not re-admitted (16 vs 15 years,  $p=0.52$ ). Of those re-admitted, 13 children underwent an invasive procedure which are detailed in table 1. This gives an overall return to theatre rate within initial admission or within 30 days of 11.0% (38/346).

### **3.5 Stoma reversal during childhood**

In those who underwent stoma reversal prior to 18 years of age ( $n=165/346$ , 48%), these took place at a median of 105 (66-161) days post IPAA formation.

## **4 Discussion**

This study reports temporal trends in paediatric IPAA surgery in England along with outcomes and highlights the role of SPS and GS in performing these procedures. IPAA for paediatric UC is undertaken infrequently and mainly in older teenagers. Just over half of children underwent colectomy and IPAA concurrently and it appears that the number of cases undertaken by SPS, in relation to GS, is increasing.

Given the infrequency of IPAA surgery in children, there are limited studies that look beyond single centre practice and, to our knowledge, this is the largest multicentre study on this topic.<sup>(15)</sup> Despite this there were as few as 12 IPAA formations nationally in one year of the study. Given that there are 21 SPS centres in England and assuming all centres are undertaking IPAA surgery the number of cases per centre per year is only 0.4. This is much lower than the 10 case minimum suggested in the European Crohn's and Colitis Organisation and European Society of Paediatric Gastroenterology, Hepatology and Nutrition guideline from 2018. In reality, the cases are unlikely to be uniformly distributed amongst centres meaning that some centres may be achieving the suggested minimum whilst other centres may not offer IPAA at all. As the study progressed, the specialty undertaking this surgery shifted from GS to SPS. This may be due to a variety of factors such including improved provision of paediatric gastroenterology and retirement of general surgeons with paediatric expertise.<sup>(12)</sup>

Given the complexity of IPAA surgery, combined with the unfavourable conditions for tissue healing that UC creates, the post-operative complication rate is significant and the short-term return to theatre rate in this current study was 11%. This has been reported at 6.7-33.3% with a mean of 20.9% on meta-analysis previously.<sup>(16)</sup> Systematic review of all paediatric IPAA has shown that surgical site infection, anastomotic leak and small bowel obstruction were seen in 10%, 11% and 14% children respectively undergoing this surgery.<sup>(15)</sup> Anastomotic leak, causing pelvic sepsis, is a particularly significant complication as pouch loss will occur in a third of these children. One of the lowest rates of anastomotic leak (1.6%) was reported in a single centre series of hand-sutured anastomoses, accepting that there was no comparative group.<sup>(17)</sup> It is also clear that post-operative complications are more frequent in IPAA surgery for UC compared to other indications.<sup>(17)</sup>

Length of stay can be used as a surrogate to determine whether a post-operative course was complicated or un-complicated. The median length of stay, calculated each year, almost halved in this study when comparing 1997 (11 days) to 2014 (6 days). A study from the United States also reported



length of stay and found the median to be 7 days in those without a major complication but this increased to 13.5 days when a major complication occurred.(18) Another factor that has previously been recognised to reduce length of stay is use of laparoscopy versus open procedures.(18) Given, the number of laparoscopic procedures identified in this study was low (15.9%) it has not been possible to explore this association in the current dataset. A clear benefit of using administrative data at population level is that it allows an accurate assessment of re-admission regardless of which hospital a child re-presented to. This is particularly important when considering the location of specialist paediatric surgical centres in England. The re-admission rate (16.8%) seen in this study is almost half the mean rate (28.9%) calculated from studies analysed with meta-analysis.(16) This low rate is reassuring and can be used by centres undertaking IPAA to benchmark their performance.

IPAA is commonly undertaken in either two (IPAA and colectomy followed by ileostomy reversal) or three (IPAA followed by colectomy followed by ileostomy reversal) stages. The number of stages is primarily the surgeons' choice but will be influenced by patient specific factors such as control of UC and steroid usage.(17)(1)(1) In this study, just under half of children had a two-stage procedure with the remainder having a three-stage approach. In a retrospective observational study from the United States, just over 40% children had a two stage approach and the outcomes were similar between both groups.(19) This suggests that surgeons were appropriately selecting whether children should have a two- or three-stage IPAA given the retrospective nature of the study. Randomisation would be required to test whether there is an advantage to either approach, however the practicalities of this, given the heterogeneity in these children, makes such a study incredibly challenging to design.

The use of laparoscopy was low in this study (15.9%), perhaps due to its low use generally at the beginning of the study, or the perception by surgeons that there is no benefit to its use in IPAA surgery. A retrospective study of laparoscopy in IPAA from the United States found that this technique was utilised in two-thirds of cases and was not associated with an increase in major adverse outcomes.(18) Moreover, it was associated with fewer minor complications and reduced length of stay. Robotic

surgery has also been proposed for IPAA surgery however there are few paediatric surgeons with regular robotic practice.(20)

This study is limited by its use of administrative data but benefits from a population based approach for an infrequently carried-out procedure. Although, it is possible that complications may have been missed if a patient re-presented outside of England. The outcomes in this study are short term only and focus on the immediate post-operative period, reports of longer term outcomes are available however there is a clear need for population level reporting of these outcomes going forward.(15,16,21) An approach to this might involve obtaining HES data to follow these children long into adulthood. Complications have only been identified when associated with a repeat procedure, identified by the OPCS-4 code. As with most studies of IPAA, patient numbers limit the ability to undertake statistical analysis, but the focus of this report is to provide clear outcomes that can be used for surgeon benchmarking. It is also important to recognise the limitations of HES data particularly prior to 2001 where it is known that missed or inaccurate coding was more common.(22)

## **5 Conclusion**

This population based study of paediatric IPAA surgery for UC has described for the first time which patients are undergoing surgery and by whom. The outcomes including length of stay, readmission rate and re-intervention rate within 30 days are comparable to other studies and provide an opportunity for individual centre and surgeon benchmarking. This is essential to ensure that outcomes keep improving for an infrequent procedure such as IPAA.

## **References**

1. Baillie CT, Smith JA. Surgical strategies in paediatric inflammatory bowel disease. Vol. 21, World Journal of Gastroenterology. WJG Press; 2015. p. 6101–16.
2. Ashton JJ, Cullen M, Afzal NA, Coelho T, Batra A, Beattie RM. Is the incidence of paediatric inflammatory bowel disease still increasing? Archives of Disease in Childhood. 2018;

3. Ashton JJ, Coelho T, Ennis S, Vadgama B, Batra A, Afzal NA, et al. Endoscopic versus histological disease extent at presentation of paediatric inflammatory bowel disease. *Journal of Pediatric Gastroenterology and Nutrition*. 2016;62(2).
4. Jakobsen C, Bartek J, Wewer V, Vind I, Munkholm P, Groen R, et al. Differences in phenotype and disease course in adult and paediatric inflammatory bowel disease--a population-based study. *Alimentary pharmacology & therapeutics*. 2011 Nov;34(10):1217–24.
5. Ashton JJ, Versteegh HP, Batra A, Afzal NA, King A, Griffiths DM, et al. Colectomy in pediatric ulcerative colitis: A single center experience of indications, outcomes, and complications. *J Pediatr Surg*. 2016;51(2):277–81.
6. Krauthammer A, Tzivnikos C, Assa A, Miele E, Strisciuglio C, Urlep D, et al. Long-term Outcomes of Paediatric Patients Admitted with Acute Severe Colitis - A Multicenter Study from the Paediatric IBD Porto Group of ESPGHAN. *Journal of Crohn's & colitis*. 2019 May 23;
7. Mattioli G, Castagnetti M, Gandullia P, Torrente F, Jasonni V, Barabino A V. Stapled restorative proctocolectomy in children with refractory ulcerative colitis. *J Pediatr Surg*. 2005;40(11):1773–9.
8. Pakarinen MP, Natunen J, Ashorn M, Koivusalo A, Turunen P, Rintala RJ, et al. Long-term outcomes of restorative proctocolectomy in children with ulcerative colitis. *Pediatrics*. 2009;123(5):1377–82.
9. Alexander F, Sarigol S, DiFiore J, Stallion A, Cotman K, Clark H, et al. Fate of the pouch in 151 pediatric patients after ileal pouch anal anastomosis. In: *Journal of Pediatric Surgery*. W.B. Saunders; 2003. p. 78–82.
10. Koivusalo A, Pakarinen MP, Rintala RJ. Surgical complications in relation to functional outcomes after ileoanal anastomosis in pediatric patients with ulcerative colitis. *Journal of Pediatric Surgery*. 2007 Feb;42(2):290–5.
11. Kelley-Quon LI, Tseng CH, Jen HC, Ziring DA, Shew SB. Postoperative complications and health care use in children undergoing surgery for ulcerative colitis. *J Pediatr Surg*. 2012;47(11):2063–70.
12. Ashton JJ, Paul T, Spray C, Devadason D, Stanton M. Current practice in UK centres providing surgery in paediatric inflammatory bowel disease - a survey from the BSPGHAN IBD working group. *Journal of Pediatric Gastroenterology and Nutrition*. 2019;68:1–1243.
13. Chang S, Shen B, Remzi F. When not to pouch: Important considerations for patient selection for ileal pouch-anal anastomosis. Vol. 13, *Gastroenterology and Hepatology*. Gastro-Hep Communications, Inc.; 2017. p. 466–75.
14. Rajaratnam SG, Eglinton TW, Hider P, Fearnhead NS. Impact of ileal pouch-anal anastomosis on female fertility: Meta-analysis and systematic review. Vol. 26, *International Journal of Colorectal Disease*. 2011. p. 1365–74.
15. Lightner AL, Alsughayer A, Wang Z, McKenna NP, Seisa MO, Moir C. Short- And long-term outcomes after ileal pouch anal anastomosis in pediatric patients: A systematic review. *Inflammatory Bowel Diseases*. 2019 Jun 18;25(7):1152–68.
16. Drews JD, Onwuka EA, Fisher JG, Huntington JT, Dutkiewicz M, Nogalska A, et al. Complications after proctocolectomy and ileal pouch-anal anastomosis in pediatric patients: A systematic review. Vol. 54, *Journal of Pediatric Surgery*. W.B. Saunders; 2019. p. 1331–9.

17. Maruthachalam K, Bunn SK, Jaffray B. Complications following restorative proctocolectomy in children. *Journal of Pediatric Surgery*. 2011 Feb;46(2):336–41.
18. McKenna NP, Potter DD, Bews KA, Glasgow AE, Mathis KL, Habermann EB. Ileal-pouch anal anastomosis in pediatric NSQIP: Does a laparoscopic approach reduce complications and length of stay? *Journal of Pediatric Surgery*. 2019 Jan 1;54(1):112–7.
19. Pruitt LCC, Bucher BT, Allen CMC, Short SS. Early ileal pouch anal anastomosis for ulcerative colitis in children: Similar outcome to delayed pouch construction despite higher comorbidity. *Journal of Pediatric Surgery*. 2021 Feb 1;56(2):245–9.
20. Lightner AL, Kelley SR, Larson DW. Robotic platform for an IPAA. *Diseases of the Colon and Rectum*. 2018;61(7):869–74.
21. Potter DD, Moir CR, Day CN, Harmsen WS, Pemberton JH. Fertility and Sexual Function in Women Following Pediatric Ileal Pouch-Anal Anastomosis. *Journal of Pediatric Surgery*. 2020 Jan 1;55(1):59–62.
22. Boyd A., Cornish R., Johnson L., Simmonds S., Syddall H., Westbury L., Cooper C., Macleod J. *Understanding Hospital Episode Statistics (HES)*. London, UK: CLOSER; 2017. Available from: <https://www.closer.ac.uk/wp-content/uploads/CLOSER-resource-understanding-hospital-episode-statistics-2018.pdf>

### **Table and figure legends**

Table 1 – Complications requiring repeat procedure within initial admission or during readmission within 30 days of IPAA. \*Multiple procedures were carried out in some children hence the column totals are greater than number of children.

Figure 1 – Pouch Surgery by Year and Speciality,  $p=0.0003$ .

Figure 2 – Median length of stay per year following pouch surgery,  $p<0.0001$ ,  $R^2=0.66$ .

**Table 1**

|   | Initial admission (n=25*) | Re-admission within 30 days (n=13*) |
|---|---------------------------|-------------------------------------|
| Revision or excision of anastomosis, n                        | 13                        | 3                                   |
| Laparoscopy/ laparotomy with further procedure unspecified, n | 8                         | 0                                   |
| Drainage of collection using interventional radiology, n      | 5                         | 2                                   |
| Surgical drainage of collection, n                            | 0                         | 4                                   |
| Perianal procedure including examination under anaesthesia, n | 0                         | 5                                   |
| Adhesiolysis, n   | 3                         | 2                                   |

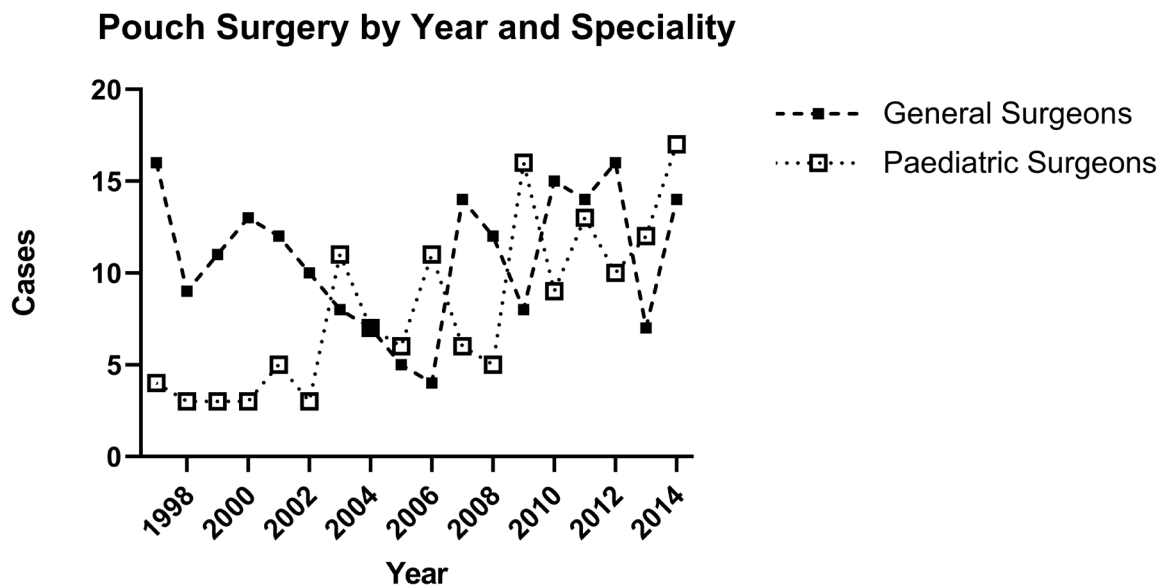


Figure 1.

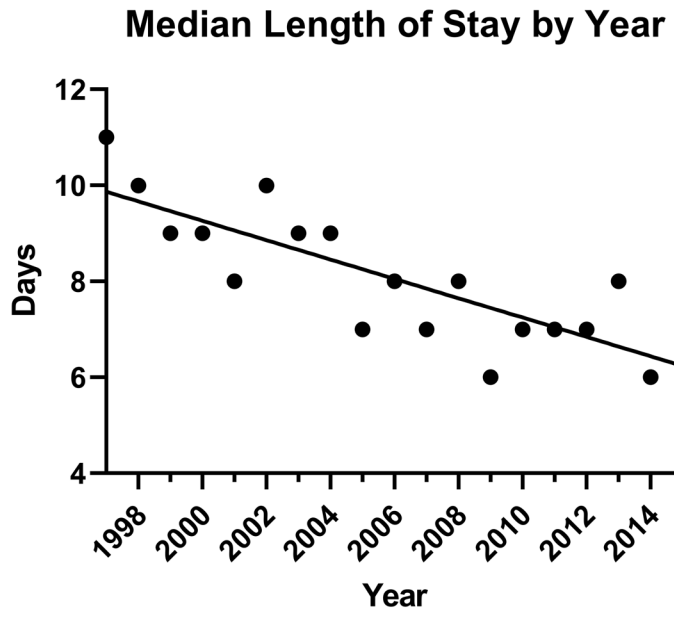


Figure 2.