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Research Article

Complications and functional outcomes after restorative proctocolectomy with ileal pouch–anal anastomosis in children: A single-center experience

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ABSTRACT

BACKGROUND: Restorative proctocolectomy with ileal pouch–anal anastomosis is a prominent treatment for adult patients with ulcerative colitis and familial adenomatous polyposis, with satisfactory functional outcomes. In literature, that technique in pediatric practice is described; however, in the domestic literature, there is no mention of ileal pouch–anal anastomosis in children.

AIM: This study aimed to examine the outcomes of ileal pouch–anal anastomosis in pediatric patients.

MATERIALS AND METHODS: The study comprised 33 patients with an ileal pouch–anal anastomosis between January 2019 and June 2023. At the time of the ileal pouch–anal anastomosis, the average age was 13 (±5) yr. Patients were followed for an average of 17 (±14) months.

RESULTS: Patients with ulcerative colitis underwent three-stage surgical interventions more often than patients with another diagnosis (90% vs. 4%, $p < 0.0001$), and the mean duration of surgery in ulcerative colitis patients was shorter than in patients with polyposis syndromes or total agangliosis: 173 (±57) min versus 280 (±73) min. Late complications were reported in five (15%) patients undergoing ileal pouch–anal anastomosis. After the ileal pouch–anal anastomosis, analysis of patient questionnaires revealed that children had satisfactory functional results.

CONCLUSIONS: Several encouraging studies have confirmed good functional outcomes after ileal pouch–anal anastomosis. Our findings suggest that ileal pouch–anal anastomosis in children is associated with favorable results

Keywords: proctocolectomy restorative; familial adenomatous polyposis; ulcerative colitis; total colonic aganglionosis; children.

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Научная статья

Осложнения и функциональные результаты после формирования тазовых тонкокишечных резервуаров у детей. Опыт одного центра

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АННОТАЦИЯ

Актуальность. Колопроктэктомия с формированием тазового тонкокишечного резервуара занимает лидирующую позицию при лечении взрослых пациентов с язвенным колитом и аденоматозным полипозным синдромом и имеет удовлетворительные функциональные результаты. В зарубежной литературе описано применение указанной методики и в педиатрической практике, однако в отечественной литературе упоминание о формировании тазовых тонкокишечных резервуаров у детей отсутствует.

Цель — анализ собственных результатов формирования тонкокишечных резервуаров у пациентов детского возраста.

Материалы и методы. В ретроспективное исследование включено 33 пациента, которым в период с января 2019 г. по июнь 2023 г. проведено формирование тазового тонкокишечного резервуара. Средний возраст пациентов на момент формирования тонкокишечного резервуара составил 13 (±5) лет. Медиана наблюдения за пациентами — 17 ± 14 мес.

Результаты. Пациентам с язвенным колитом чаще, чем пациентам с другими диагнозами проводились 3-этапные оперативные вмешательства (90 % против 4 %, $p < 0{,}0001$), в связи с этим среднее время формирования тазового тонкокишечного резервуара у пациентов с язвенным колитом короче, чем у пациентов с полипозными синдромами или тотальным аганглиозом — 173 (±57) мин против 280 (±73) мин. Из 33 пациентов с тонкокишечным резервуаром поздние осложнения зарегистрированы у 5 (15 %) пациентов. Анализ анкет пациентов показал, что дети в нашем исследовании имеют удовлетворительные функциональные результаты после проведенной операции.

Заключение. Мы обладаем достаточным количеством обнадеживающих исследований, подтверждающих хорошие функциональные исходы после формирования тазового тонкокишечного резервуара, в том числе и у детей. Наше исследование показало, что формирование тонкокишечного резервуара у детей в нашей клинике сопряжено с удовлетворительными результатами.

Ключевые слова: тазовый тонкокишечный резервуар; аденоматозный полипозный синдром; язвенный колит; тотальный аганглиоз; дети.

Как цитировать

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儿童盆腔小肠贮液器形成后的并发症和功能结果。单个中心的经验

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简评

论证。在溃疡性结肠炎和腺瘤性息肉病综合征成人患者的治疗中，结肠肛门切除术伴有盆腔小肠贮液器的形成占据主导地位，并取得令人满意的功能效果。国外文献也介绍这种技术在儿科临床中的应用。然而，国内文献中并未提及儿童盆腔小肠贮液器的形成。

该研究的目的是对我们自己在儿科患者小肠贮液器形成方面取得的成果进行分析。

材料与方法。本回顾性研究共纳入了33名患者。这些患者在2019年1月至2023年6月期间接受了盆腔小肠贮液器成形术。小肠贮液器形成时患者的中位年龄为13（±5）岁。患者的中位随访时间为17±14个月。

结果。与其他诊断的患者相比，溃疡性结肠炎患者更有可能接受三阶段手术干预（4%对90%，$p < 0.0001$）。因此，与息肉病综合征或先天性巨结肠症患者相比，对于溃疡性结肠炎患者来说，盆腔小肠贮液器形成的平均时间更低（280（±73）分钟对173（±57）分钟）。在33名小肠贮液器患者中，有5名（15%）患者出现了晚期并发症。对患者问卷的分析表明了，本研究中的患儿术后功能效果令人满意。

结论。我们有大量令人鼓舞的研究。这些研究证实，盆腔小肠贮液器成形术后的功能效果良好（包括儿童在内）。我们的研究表明了，在我们的诊所中，为儿童形成小肠贮液器的效果令人满意。

关键词：盆腔小肠贮液器；腺瘤性息肉病综合征；溃疡性结肠炎；先天性巨结肠症；儿童。

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INTRODUCTION

Restorative proctocolectomy with ileal pouch–anal anastomosis (IPAA) has taken a leading position in the treatment of ulcerative colitis (UC) and familial adenomatous polyposis (FAP) in adults because of satisfactory functional results [1, 2]. This technique is gaining popularity in pediatric practice, including the treatment of UC, FAP, and total colonic agangliosis (TCA) [3–5].

Historically, IPAA formation was proposed to restore anal defecation in patients after proctocolectomy in UC, a chronic autoimmune disease of the colonic mucosa. The prevalence of UC in children and adolescents ranges from 31 to 75 per 100,000, with an annual increase in incidence [6]. Approximately 20% of UC cases manifest in childhood, which have a more severe course than adults. In addition, nearly 13% of children require colectomy within 3 years of diagnosis [7, 8]. Most UC cases are treated conservatively. The need for surgery arises when therapy is ineffective, steroid dependence and resistance develop, and intestinal complications such as toxic megacolon with colonic perforation or intestinal bleeding occur [9].

FAP is a rare an autosomal dominant disease characterized by the development of hundreds of thousands of colorectal adenomas and a 100% risk for colorectal cancer without timely radical surgical intervention. FAP is caused by mutation in the *APC* gene and occurs in 1 of 10,000 newborns. FAP is the second most common cause of colorectal cancer among hereditary syndromes and is responsible for 1% of all colorectal cancers [10]. The disease has a 50% chance of being inherited from a parent, and 20%–30% of cases can develop so-called new germinal mutations, or *de novo* mutations, characterized by the emergence of the disease in a person with no family history of FAP. The only treatment option for FAP is proctocolectomy [4].

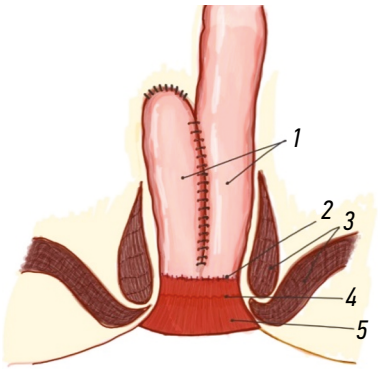

Figure. Schematic view of J-pouch: *1* — J-pouch; *2* — ileal pouch–anal anastomosis; *3* — anal sphincter; *4* — linea dentate; and *5* — anal canal

Рисунок. Схематичное изображение тазового тонкокишечного J-резервуара. 1 — тазовый тонкокишечный J-резервуар из подвздошной кишки; 2 — резервуаро-анальный анастомоз; 3 — сфинктерный аппарат; 4 — зубчатая линия; 5 — анальный канал

TCA is a rare severe congenital anomaly, which occurs in 1 per 50–100,000 newborns. TCA has a high mortality rate (up to 10%) because of the development of intestinal obstruction, Hirschprung-associated enterocolitis, and genetic syndromes that aggravate the overall condition of the patients [5]. Currently, straight ileoanal anastomosis is the main method of the surgical correction of this malformation, which often leads to unsatisfactory functional results and dictate the need to search for new solutions.

Despite the widespread introduction of pouch surgery globally, IPAA is associated with a high incidence of postoperative complications, which is reflected in many foreign studies [1, 11, 12]. In the Russian literature, the analysis of complications in adult patients after IPAA in patients with UC was detailed in the study by S.I. Achkasov et al. [13]; however, this surgery was not mentioned in pediatric patients.

This *study aimed* to analyze the results of proctocolectomy with IPAA in pediatric patients.

MATERIALS AND METHODS

A retrospective study was conducted, including 33 patients aged <18 years who restorative proctocolectomy with ileal pouches between January 2019 and June 2023.

Brief description of surgical technique

After proctocolectomy (two-stage procedures), pouch formation was started from the distal parts of the ileum. To ensure adequate mobility of the small intestine and its mesentery for subsequent adequate relegation to the small pelvis, the small intestine mesentery was mobilized from the retroperitoneal space. The J-shaped configuration of the pouch was achieved by folding the terminal ileum into a J-shape with a total length of 12–15 cm. An incision was made along the antimesenteric margin for subsequent insertion of the linear stapling apparatus, and two subsequent suturing of two adjacent sections of intestine were performed. Then, the pouch was lowered to the pelvis under visualization to avoid pouch torsion, followed by IPAA (hardware or manual), most commonly at a height of 1–2 cm above the dentate line (Figure). The descriptive characterization of the patients and the studied factors is presented in Table 1.

All patients with IPAA underwent a comprehensive evaluation before prophylactic ileostomy closure, particularly clinical examination (inspection and rectal examination to assess the condition of the anastomosis), contrast radiography of the pouch (pouchography) to exclude suture and anastomosis leakage, and endoscopic examination of the pouch to assess the condition of the pouch mucosa, patency of the anastomosis, and condition of the anal canal.

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Table 1. Descriptive characteristics of patients who underwent the restorative proctocolectomy with ileal pouch–anal anastomosis, *n* = 33 (100 %)

Таблица 1. Описательная характеристика пациентов, перенесших формирование тазового тонкокишечного резервуара, *n* = 33 (100 %)

Factors	Significance
Sex, m/f	17/16
Mean age at pouch construction (±SD), years	13 (±5)
Body mass index at pouch construction, median [25th and 75th percentiles]	17.1 [16–20.6]
Diagnosis:	
Familial adenomatous polyposis	20 (61%)
Ulcerative colitis	10 (30%)
Total colonic agangliosis	2 (6%)
Juvenile polyposis syndrome	1 (3%)
Mean duration of ulcerative colitis before ileal pouch–anal anastomosis (±SD), months	58 (±37)
Mean time from colectomy to ileal pouch–anal anastomosis (for patients with ulcerative colitis) (±SD), months	16 (±10)
Preoperative use of steroids (for patients with ulcerative colitis), *n*	10 (100%)
Preoperative use of immunosuppressive therapy (for patients with ulcerative colitis), *n*	8 (80%)
Preoperative use of biologic therapy (for patients with ulcerative colitis), *n*	9 (90%)
Associated pathology:	
Papillary thyroid cancer	1
Overweight (BMI >25)	2
Grade I obesity (BMI 31)	2
Type 1 diabetes mellitus	1
Pulmonary artery stenosis	1
Body weight deficiency (<3 percentile), familial adenomatous polyposis/ulcerative colitis	7 (21%) 5/2
Mean hemoglobin at the time of pouch formation (±SD), g/L	123 (±16)
Mean hemotocrit at the time of pouch formation (±SD), %	38 (±4)
Median white blood cell count at the time of pouch formation, ×10^{9}/L	6.3 (5.6–8.3)
Platelet count at the time of pouch formation, median [25th to 75th percentiles], ×10^{9}/L	332 [279–444]
Mean albumin at the time of pouch formation (±SD), g/L	41 (±3)
C-reactive protein at the time of pouch formation, median [25th to 75th percentiles], mg/L	0.8 [0,3–3.4]
Surgical access:	
Laparoscopic	18
Open (until 2020)	15
Pouch–anal anastomosis technique:	
Handsewn	22
Stapled	11
Mean procedure time (±SD), min	247 (±84)
Length of stay median [25th–75th percentiles], days	12 [10–14]

Statistical analisis

Available data were extracted from paper and electronic patient records and entered into an Excel spreadsheet. Data analysis was performed using GraphPad Prism version 9.3.1 (GraphPad Software, USA). Descriptive statistics were performed: after estimating the distribution (normal or non-normal), quantitative indicators were described, indicating the median (with interquartile range) for parameters with non-normal distribution and the mean (with standard deviation) for those with normal distribution. Sub-analyses were performed to compare the characteristics of surgical intervention according to diagnosis (UC, FAP, and TCA) using the Mann–Whitney test. Between-groups differences were considered statistically significant at $p < 0.05$.

RESULTS

Immediate results

In most cases of FAP and TCA, a two-stage surgery was performed, proctocolectomy with IPAA and loop ileostomy followed by intestinal stoma closure. Patients with UC were statistically significantly more likely to undergo a three-stage procedure (colectomy, proctectomy and ileostomy followed by ileostomy closure) than patients with other diagnoses (90% vs. 4%, $p < 0.0001$). Colectomy was indicated for patients with UC who had ineffective medical therapy (9) and colonic perforation (1). The mean time to reconstructive surgery (IPAA formation) from the time of colectomy in three-stage procedures was 16 (±10) months. The mean duration of the pouch–anal anastomosis in patients with UC is shorter than that in patients with polyposis syndromes and TCA, i.e., 173 (±57) min vs. 280 min, because of earlier colectomy in patients with UC. The mean duration of laparoscopic procedures was longer than that open procedures, i.e., 277 (±96) vs. 212 (±51) min ($p = 0.02$).

The mean follow-up following IPAA was 17 ± 14 months. Of these, the ileostomy was closed in 28 (85%) patients as of June 2023. The median time following stoma closure was 7.5 (3–12) months.

Early postoperative complications were observed in 3 (9%) patients. Of these, 2 (6%) children with infravesical obstruction required a transurethral bladder neck incision with complete restoration of urination. According to catamnestic observation data, one patient had no urinary incontinence. According to the ultrasound of the urinary organs, no residual urine was found in the bladder, and the collecting system of the kidneys was not enlarged (duration of catamnesis was 28 months). The second patient was lost to follow-up when reaching the age of 18. One patient (3%) experienced anastomotic leakage with abscess, which required antibiotic therapy followed by secondary sutures. At the control visit, the anastomosis was found to be intact, and the ileostomy was closed after 4 months.

Long-term results

Late complications were reported in 5 (15%) of the 33 patients with IPAA. In 2 (6%) patients with FAP, pouchitis (inflammatory changes in the mucosa of the small intestinal pouch) was diagnosed by endoscopy 8 months after stoma closure, requiring antibacterial and anti-inflammatory therapy. Small bowel obstruction occurred in 2 (6%) patients following open (1) and laparoscopic (1) surgeries. These patients underwent surgery laparotomy. One patient (3%) with FAP required dissection of the anastomotic stricture 6 months following stoma closure because of severe stenosis of the pouch–anal anastomotic area. Constriction of the pouch–anal anastomosis was diagnosed in 12 (36%) patients. All cases required a single anastomotic bougie during routine endoscopic inspection of the pouch, which did not delay the timing of ileostomy closure.

Functional results

A survey of patients who underwent reconstructive surgery with ileostomy closure was conducted to assess functional outcomes. A questionnaire (pouch functional score) created by leading pouch surgeons R.E. Lovegrove and V.W. Fazio, published in the *British Journal of Surgery Society* in 2010 and validated in adult patients with IPAA [14], was used. Owing to the lack of validated scales to assess functional outcomes in children with IPAA, a survey, based on the pouch functional score and experience, of the main parameters characterizing functional outcomes in children with IPAA was independently performed. Responses were received from 18 patients. Indicators such as daytime stool frequency, presence and frequency of nocturnal defecation, daytime and nocturnal fecal incontinence, need for day/night pads, episodes of perianal dermatitis, and patients' return to sports were analyzed. The results of the survey are summarized in Table 2.

Table 2. Functional outcomes of children after restorative proctocolectomy with ileal pouch–anal anastomosis, *n* = 33 (100 %)
Таблица 2. Функциональные результаты детей после формирования тазового тонкокишечного резервуара, *n* = 33 (100 %)

Parameters	Significance
Mean daytime stool frequency (±SD)	5 (±2)
Mean nocturnal stool frequency (±SD)	1 (±1)
Daytime fecal incontinence, *n*	6 (33%)
Nocturnal fecal incontinence, *n*	3 (17%)
Need for daytime pad use, *n*	6 (33%)
Need for nocturnal pad use, *n*	5 (28%)
Sports, *n*	6 (33%)
At least one episode of perianal dermatitis (including postoperative period), *n*	8 (44%)

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DISCUSSION

Currently, the national literature does not mention IPAA formation in children; however, foreign publications demonstrated extensive experience in performing this surgery, describing good functional results [15, 16]. According to the clinical recommendations of the Association of Coloproctologists of Russia, IPAA surgery enables controlled defecation through the anus, with a defecation frequency of 4–8 times a day in adults and an average daily volume of semiformal/liquid stool of 700 mL per day, which is considered a satisfactory result. In the analysis of the questionnaires, patients who underwent IPAA formation had a mean stool frequency of 5 (±2) times a day with the urge to defecate. Grade I–II anal incontinence and periodic nocturnal fecal incontinence were found in 6 (33%) and 3 (17%) patients, respectively [17]. The incidence of perianal dermatitis was high because every patient with at least one episode of perianal dermatitis, including the early postoperative period, was included in the study. In the long term, patients did not complain of any dermatitis or discomfort in the anal area.

Despite encouraging functional results in our observation and according to the literature, IPAA remains serious procedure with a rate of pouch-related complications as high as 75%. Several studies have analyzed complications following IPAA formation in children [18–24]. For example, a multicenter study from Japan analyzed outcomes following IPAA in 212 children with UC. Bowel intestinal obstruction (20%) and wound problems (15%) were the most common early postoperative complications. According to the authors, pouchitis occurred in 18% of the patients and fistulas related to pouch in 13% [20].

A systematic review by J.D. Drews analyzed complications after IPAA in 763 patients aged <21 years with UC and FAP. The incidence of anastomotic leakage was as high as 10% for patients with UC and 5% for patients with FAP. In patients with UC, the incidence of pouchitis varied widely (up to 76%), significantly higher than that in patients with FAP (12%). The incidence of stricture of the anastomosis reached 14%; however, in most cases, the structure was eliminated with a single anastomotic dilatation [21]. In our observation, 1 patient had rigid stenosis of the anastomosis requiring stricturoplasty, and 12 patients with anastomotic stenosis were successfully treated with a single dilatation before stoma closure.

Regarding strictures of pouch anastomosis, Diederen et al. [22] revealed the outcomes after IPAA in 445 patients (adults and children) [22]. The study found that childhood age was an independent risk factor for the stenosis of the pouch–anal anastomosis (odds ratio [OR], 4.22; 95% confidence interval [CI], 1.13–15.77; $p = 0.032$). Why the incidence of anastomotic stricture is so high in pediatric patients compared with adults remains debatable. This may be due to the diameter of the stapler, traumatization of the mucosa by the stapler head, or other peculiarities of the anastomotic technique [22]. However, the high incidence of anastomotic strictures in children has not be precisely explained.

Nyholm et al. [23] described the results of pouch–anal anastomosis in 87 patients with UC, and the incidence of postoperative complications reached 55%. The most common complication in this cohort was small intestinal obstruction (29%), which required surgical intervention. Polites et al. [24] analyzed the incidence of postoperative complications following pouch formation in 81 children with UC and FAP. The complication rate was 54% [24].

Shannon et al. analyzed the results of the IPAA in 74 patients with UC [25]. The authors found a high incidence of postoperative complications such as pouchitis (45%), stricture of the pouch anastomosis (16%), fistulas related to pouch (30%), intestinal obstruction (20%), and pouch failure (14%).

Given the significant incidence of complications following IPAA in children, factors associated with the risk of postoperative complications must be determined. Some authors have included obesity and immunosuppressive and hormonal therapy in the preoperative period [26]. The administration of prednisolone at a dose >20 mg for >6 weeks is known to increase the incidence of surgical complications [27].

Dukleska et al. [28] found an association between excess patient weight and the risk of repeated surgeries for complications (OR, 3.34; 95% CI, 1.08–10.38; $p = 0.04$). Huang et al. [29] analyzed whether the diagnosis and complications following IPAA are related. The study included 79 children with UC (62) and FAP (17). Patients with FAP were statistically significantly less likely to have pouchitis (8% vs. 49% with UC, $p = 0.009$) and less likely to be diagnosed with pouch inefficiency (0% vs. 4% in UC, $p < 0.001$) [29]. In our observation, the overall incidence of pouch-associated complications was 24%, which is comparable to published data [13]. The incidence rates of complications such as anastomotic leakage and intestinal obstruction were low at 3% and 6%, respectively.

The incidence of postoperative complications in our sample does not exceed foreign literature data. Nevertheless, the high rate of postoperative complications requires research to identifying predictors of adverse outcomes in children following IPAA formation. Undoubtedly, the question of choosing a method of anal continence repair with minimal postoperative complications and providing patients with an acceptable quality of life remains a burning issue in the practice of pediatric surgeons, which dictates the need for studies comparing these two techniques and the functional outcomes and quality of life of patients following such surgeries. Further catamenial follow-up of patients following IPAA formation will evaluate long-term outcomes such as sexual dysfunction, fertility, and pouch inefficiency.

CONCLUSIONS

IPAA is a promising trend in pediatrics and is introduced into the practice of pediatric surgeons worldwide. Published studies confirm good functional outcomes after IPAA formation in children, despite a relatively high percentage of pouch-associated complications. However, surgeries of this complexity should only be performed in specialized centers with extensive experience in such reconstructive procedures. This study showed that IPAA formation during childhood is associated with satisfactory outcomes in the patient population studied. Further studies will allow finding the optimal variant of surgical treatment for this severe category of patients, identifying predictors of complications, and analyzing the long-term functional outcomes, including the quality of life.

ADDITIONAL INFORMATION

Authors' contribution. Thereby, all authors made a substantial contribution to the conception of the study, acquisition, analysis, interpretation of data for the work, drafting and revising the article, final approval of the version to be published and agree to be accountable for all aspects of the study. The contribution of each author: L.R. Khabibullina — surgical treatment and curation of patients, conducting a literature review, collecting and analyzing literary sources, collecting and processing materials, writing the text of an article, statistical data processing; A.Yu. Razumovsky — article editing; O.V. Shcherbakova — surgical treatment of patients, article editing.

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Competing interests. The authors declare that they have no competing interests.

ДОПОЛНИТЕЛЬНАЯ ИНФОРМАЦИЯ

Вклад авторов. Все авторы внесли существенный вклад в разработку концепции, проведение исследования и подготовку статьи, прочли и одобрили финальную версию перед публикацией. Личный вклад каждого автора: Л.Р. Хабибуллина — хирургическое лечение и курация пациентов, обзор литературы, сбор и анализ литературных источников, сбор и обработка материалов, написание текста статьи, статистическая обработка данных; А.Ю. Разумовский — редактирование статьи; О.В. Щербакова — хирургическое лечение пациентов, редактирование статьи.

Источник финансирования. Авторы заявляют об отсутствии внешнего финансирования при проведении исследования и подготовке публикации.

Конфликт интересов. Авторы декларируют отсутствие явных и потенциальных конфликтов интересов, связанных с проведенным исследованием и публикацией настоящей статьи.

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