



Optimizing surgical resection of the bleeding Meckel diverticulum in children



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ABSTRACT

Purpose: Meckel diverticula containing gastric heterotopia predispose to local hyperacidity, mucosal ulceration, and gastrointestinal bleeding in children. Eradication of acid-producing oxyntic cells is performed by either of two surgical methods: segmental enterectomy including the diverticulum or diverticulectomy only.

Methods: Retrospective review of all children having surgical resection of a Meckel diverticulum at a tertiary-referral children's hospital from 2002 to 2016 was performed. Demographic data, surgical method, pathological specimens, and outcomes were evaluated.

Results: 102 children underwent surgical resection of a Meckel diverticulum during the study period. 27 (26.5%) children presented with bleeding, of which 16 (59%) had diverticulectomy only, and 11 (41%) had segmental ileal resection. All Meckel diverticula in children presenting with bleeding contained gastric heterotopia, and resection margins were free of gastric mucosa. Histologically, 19 specimens showed microscopic features of ulceration, on average 2.95 mm (SD 4.49) from the nearest gastric mucosa (range: 0–16 mm). Mean length of hospitalization after ileal resection was 4.0 days (SD 1.2) compared to 1.6 days (SD 0.9) for diverticulectomy only ($p < 0.001$), with no re-bleeding occurrences.

Conclusion: In the operative management of children having a bleeding Meckel diverticulum, diverticulectomy-only completely eradicates gastric heterotopia without increased risk of continued bleeding or complications and significantly shortens hospitalization.

Level of evidence: Treatment Study: Level III.

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Meckel diverticulum is an outpouching of the mid-ileum that develops as a result of an incompletely obliterated omphalomesenteric, or vitelline, duct [1]. The prenatal yolk stalk investing the omphalomesenteric duct typically regresses between the fifth and seventh weeks of fetal life. Errors in the coordination of this yolk stalk regression can lead to variable entrapment of primordial intestinal tissue and ileal anomalies. Indeed, an omphalomesenteric duct will persist in approximately 2% of the population forming a true diverticulum located typically 45–60 cm proximal to the ileocecal valve [2–4]. Given its relative commonality, Johann Meckel in 1809 described this congenital diverticulum as the most prevalent anomaly of the alimentary tract [5,6].

The early embryologic origin of the omphalomesenteric duct, coupled with the short length of the intestinal tract at this stage of development, implies that the duct lining will contain multipotent stem cells normally fated to become a variety of different digestive tissues but which become trapped in this vitelline remnant. Specifically, a Meckel diverticulum can contain ectopic gastric and duodenal mucosa and more rarely rests of

pancreatic tissue. As a result, it has been estimated that 4–6% of individuals with a Meckel diverticulum will become symptomatic, presenting most commonly with bleeding, obstruction, diverticulitis, or perforation [3,7]. Specifically, Meckel diverticulum is the most common cause of small intestinal bleeding in children, occurring in a reported 31% of children with a symptomatic Meckel diverticulum [8,9].

Pathologically, a Meckel diverticulum containing gastric heterotopia predisposes to local hyperacidity, mucosal ulceration, and gastrointestinal bleeding in children. Eradication of the heterotopic, acid-producing oxyntic cells is performed by either of two surgical methods: segmental enterectomy including the diverticulum or diverticulectomy only. Advocates of small bowel resection for a bleeding Meckel diverticulum may choose this therapy because of concern for heterotopic gastric mucosa at the base or within the ileum or to incorporate any potential ileal ulceration within the resection specimen [10–12]. Alternatively, advocates of diverticulectomy-only contend that the ulcerated intestinal mucosa is immediately adjacent to the heterotopic gastric mucosa, which itself originates in the diverticulum, and that removal of this acid-producing source is sufficient to prevent further bleeding [13]. Diverticulectomy-only has also been reported as performed in both adults and children via both an open or laparoscopic approach, and the latter may confer additional convalescent advantages [14–16].

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Much of the controversy regarding the optimal surgical therapy for the bleeding Meckel diverticulum is because of the paucity of published data on the precise location of the ulceration and its proximity to the heterotopic gastric mucosa. Information on the location of the acid-producing cells and actual source of bleeding could assist in guiding more streamlined and higher quality surgical care [17]. Our overarching hypothesis states that the acid concentration is greatest immediately adjacent to the gastric mucosa and is buffered rapidly with increasing distance, thereby predisposing the immediately adjacent intestinal-like mucosa within the diverticulum to be the site of acid-induced ulceration. Therefore, we sought to test the specific hypothesis that diverticulectomy-only is sufficient to eliminate the ectopic acid source, ulcer, and bleeding symptoms.

1. Methods

We reviewed retrospectively the outcomes of all children having surgical resection of a Meckel diverticulum at a tertiary-referral children's hospital between 2002 and 2016 to evaluate if diverticulectomy-only is sufficient treatment for bleeding Meckel diverticula. We also re-reviewed all original Meckel specimens resected for bleeding to document the presence of heterotopic gastric mucosa, measure the distance of intestinal ulceration from the nearest edge of oxyntic cells, and assess resection margins for the absence of gastric mucosa.

1.1. Subjects and setting

The study population consisted of all children treated for a Meckel diverticulum at the Monroe Carell, Jr. Children's Hospital of the Vanderbilt University Medical Center (VUMC) in Nashville, TN from January 1, 2002 to August 31, 2016. All records containing an ICD-9 diagnosis code of 751.0 or ICD10 code Q43.0 (i.e., Meckel diverticulum) or C17.3 (i.e., Meckel diverticulum, malignant) at 18 years of age or younger were ascertained. Charts were reviewed manually by a single reviewer (JRR) with a second reviewer (HNL) involved to address uncertainty. Children were excluded if there was no mention in the attending pediatric surgeon's operative report of a visible diverticulum extruding from the ileum or pathology reports. Cases of heterotopic gastric mucosa occurring in the absence of a Meckel diverticulum were excluded. The institutional review board at VUMC approved the study and waived requirement for obtaining informed consent.

1.2. Categorization of presenting characteristics

All patient records were reviewed to ascertain demographic data, including age at presentation, race, and ethnicity. Records were reviewed to determine the clinical symptoms preceding operative management for the Meckel diverticulum. Bleeding was defined as bloody stools with or without a declining hemoglobin or hematocrit. Operative reports were reviewed to determine the method of surgical resection and classified as open diverticulectomy, laparoscopic diverticulectomy (Fig. 1), or segmental ileal resection. Diverticulectomy-only was defined as resection of the diverticulum alone, without performing a resection of a circumferential portion of the small intestine (Fig. 1). Any missing data were categorized as unknown.

1.3. Assessment of clinical outcomes

The main outcomes of interest included adverse events and length of hospital stay post-operatively. Adverse events were predefined as continued bleeding post-operatively (need for post-operative transfusion or continued blood in stool), surgical site infections, post-operative emergency department visits, hospital readmissions, respiratory distress, bacteremia, pneumonia, urinary tract infection, additional operative or interventional radiology procedures, or other complications of treatment. All adverse events documented in the electronic medical

record within 90 days of surgical resection were included in analysis. Length of hospital stay after surgery was measured as the time from surgery to discharge order. Categorical outcomes were compared using Fisher exact test and continuous variables with t-tests. All analyses were conducted in R version 3.0.1 [18].

1.4. Analysis of pathology specimens

Pathology reports of all patients having resection of a Meckel diverticulum were reviewed to determine the presence or absence of heterotopic tissue, including gastric and pancreatic. Of patients who presented with a bleeding Meckel diverticulum, hematoxylin and eosin (H&E) stained slides of the original pathology specimens were retrieved for detailed histologic re-analysis. Each specimen was analyzed specifically for the presence and location of gastric heterotopia. Pathologic margins were assessed further for the presence of gastric heterotopia. All available H&E slides from each specimen were analyzed in detail for the presence of mucosal ulceration or erosion (Fig. 2). If ulceration or erosion was present within the sampled specimen, the distance from gastric heterotopia to ulceration was measured microscopically in millimeters (mm). As heterotopic pancreatic tissue was unlikely to contribute to bleeding or mucosal ulceration, we did not analyze the relationship of pancreatic heterotopia to ulceration or gastric heterotopia.

2. Results

2.1. Demographic characteristics

A total of 102 patients underwent surgical resection of a Meckel diverticulum during the study period. Clinical and demographic characteristics of the entire cohort are presented in Table 1. The study population predominantly consisted of children who were white (90%) and male (75%). Overall, the two most common presenting symptoms were obstruction ($n = 32$) and bleeding ($n = 27$), respectively. The majority of patients ($n = 70$; 69%) underwent diverticulectomy only, with 40 children having an open diverticulectomy and 30 laparoscopic diverticulectomy. For the entire cohort, and regardless of surgical indication, 60 children (59%) had gastric heterotopia, and 7 children (7%) had pancreatic heterotopia within the diverticulum. Those children who developed bleeding were not significantly different in age compared to other symptomatic presentations of a Meckel diverticulum (7.0 versus 8.3 years; $p = 0.39$).

2.2. Clinical outcomes of all children undergoing surgery for Meckel diverticulum

Of all children who underwent surgical resection of a Meckel diverticulum for any reason, 7 of 102 (6.9%) had a post-operative complication, none of which occurred in the bleeding sub-group. The majority (4 of 7 children) who developed a complication underwent resection of a Meckel diverticulum discovered incidentally at time of surgery for another indication (e.g., malrotation or necrotizing enterocolitis). Mean age of children who had a complication after undergoing surgical resection of an incidentally discovered Meckel diverticulum was 1.8 years. For the entire cohort, only one death occurred, that of a newborn who was operated on for obstruction secondary to intestinal malrotation who had concomitant and complex congenital cardiac anomalies that resulted in demise of the infant on post-operative day 11. None of these complications could be directly attributed to resection of the incidentally discovered Meckel diverticulum.

The remaining 3 children who developed a post-operative complication had resection of a Meckel diverticulum for symptoms of obstruction. Of patients with obstruction, one underwent a segmental ileal resection and was the only patient in the cohort to have a post-operative anastomotic leak requiring reoperation. A single patient who underwent open diverticulectomy for obstruction secondary to

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