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A comparison of suction and full-thickness rectal biopsy in children

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ABSTRACT

Background: Rectal biopsy evaluation by an experienced pathologist is the gold standard in diagnosis of Hirschsprung's disease (HD). Although both suction rectal biopsy (SRB) and full-thickness (FTRB) rectal biopsy are performed, the ability for SRB to obtain adequate tissue in older children has been questioned. We hypothesized that SRB and FTRB yield tissue specimens of different size but are equally adequate for diagnosis.

Methods: Records of children who underwent rectal biopsy to evaluate for HD between January 2007 and July 2014 were reviewed. Volume, percent submucosa, and specimen adequacy were compared between biopsy techniques, and the effect of age on biopsy adequacy was assessed. Data were analyzed by mixed-effects models with covariate adjustment for age at biopsy and Fisher's exact test.

Results: Forty-seven children underwent a total of 58 biopsies, 45 SRB and 13 FTRB. Thirty-seven were performed before 12 mo of age, and 21 after 12 mo of age. Volume of SRB specimens was significantly smaller than FTRB across ages (14.8 ± 7.8 mm^3 *versus* 121.3 ± 13.8 mm^3, $P = 0.0001$). Percent submucosa did not differ significantly between SRB and FTRB specimens across ages ($63.8 \pm 2.7\%$ *versus* $66.5 \pm 4.3\%$, $P = 0.575$). The number of inadequate biopsies was low and not significantly different across ages ($P = 0.345$), or when comparing by biopsy method ($P = 0.689$). All biopsies were clinically diagnostic. There were no complications.

Conclusions: Tissue specimens obtained by SRB are smaller than those obtained by FTRB, especially in older children. SRB and FTRB appear equivalent in their ability to provide adequate submucosa. Differences in cost and patient satisfaction between rectal biopsy techniques must be studied to further define the best overall technique.

1. Introduction

Hirschsprung's disease (HD) is the most common congenital anomaly of colorectal function and affects approximately 1 in 5000 infants. Histologically, it is characterized by aganglionosis of varying distances in the rectum and colon, and rectal biopsy is the gold standard for diagnosis. HD is more common in males with a 4:1 male-to-female ratio, and its incidence in premature infants is thought to be low. Currently, most of the cases are identified during infancy

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leaving only rare instances where HD is diagnosed in an older child or during adulthood. In these older patients, poor stooling function is most often due to functional or idiopathic constipation [1–5]. The characteristic presentation of HD includes delayed passage of meconium, abdominal distention, and vomiting, and a contrast enema may demonstrate a transition zone from ganglionic to aganglionic bowel [6–8]. All these signs and symptoms need not be present to warrant a biopsy, as any concern for lower gastrointestinal obstruction in an infant should raise a suspicion for HD, and prompt a rectal biopsy.

A correctly executed rectal biopsy procedure should involve sampling a segment of rectal wall 2 cm proximal to the dentate line along the posterior wall of the rectum [9]. A biopsy taken too distally may collect a specimen from the physiologically aganglionic region erroneously suggesting the presence of HD, whereas a biopsy taken too proximally (i.e., 4 + cm) may miss very short segment HD [10,11]. Suction rectal biopsy (SRB) and full-thickness rectal biopsy (FTRB) are both regularly used and considered suitable methods for rectal biopsy. There is, however, a commonly held preference to favor SRB in younger children and FTRB in older children, based on a presumption that tissue specimens obtained by SRB in older children would be too small, and thus inadequate for diagnosis and likely lead to an error in diagnosis or a repeat biopsy [12–14]. We hypothesized that FTRB would produce larger volume specimens when compared with SRB specimens, and that both types of biopsy specimens would be sufficient for pathologic assessment and diagnosis.

2. Methods

All patients who underwent rectal biopsy for the evaluation of HD between January 2007 and July 2014 were identified from operative, clinical, and pathology records in this single institution, retrospective study. Medical records, operative reports, pathology reports, and final diagnosis were reviewed in each case. Patient demographic data were collected including age, gender, birth history, comorbidities, and biopsy result. Radiologic imaging was recorded. The primary outcome was specimen adequacy, defined as greater than 50% submucosa by depth. Secondary outcomes were biopsy volume, number of inadequate biopsy specimens, and ability to make a final diagnosis. For the purposes of this study and based on previously published studies, we chose to follow the consensus in the surgical literature and compare results before and after 1 year of age [3]. The institutional review board granted approval for this study (Protocol #1309012689).

2.1. Biopsy techniques

SRB was performed using rbi2 system (Aus Systems, Australia), as shown in Figure 1. Older children underwent SRB in the operating room, as FTRB was performed concomitantly during the same procedure. Younger children underwent SRB as a bedside procedure. After calibration to 300mmH_2O before use, the instrument was inserted into the rectum along the posterior wall and positioned with the cutting blade about 2 cm above the dentate line. Gentle pressure

Fig. 1 – rbi2 Suction rectal biopsy system [15] is manufactured by Aus Systems. rbi2 Is a reusable biopsy system with negative pressure manometer, and single-use tip housing the cutting blade. (Color version of figure is available online.) Photo courtesy of Dawn Nelson, Specialty Surgical Products, Inc [15].

was held by the operator against the wall and suction applied to 150mmH_2O using the manometer (or withdrawing the plunger to 3-5 mL using a 10-mL syringe) by the first assistant, before triggering the cutting blade [15]. Typically two or three SRB specimens were obtained in one session. There were no complications associated with the SRB technique.

FTRB was performed under general anesthesia in the operating room such that an anoscope or speculum could be positioned within the rectum to visualize the biopsy site directly. Under direct vision, FTRB specimens were obtained with scissors at about 2 cm from the anal verge. FTRB sites required suture closure for hemostasis. There were no complications associated with the FTRB technique.

2.2. Histology

The biopsy samples were provided to the pathology laboratory either fresh or in saline. The first two tissue samples were fixed in formalin, embedded in paraffin, serial sectioned at 5-6 microns, and stained with hematoxylin and eosin (H&E). If more than two biopsy samples were provided, one was frozen to allow additional staining with acetylcholinesterase (AChE). Although some patients had additional histochemical staining with AChE or immunohistochemical staining for calretinin, only H&E slides were used for our review. Gross specimen dimensions, mucosa depth, and submucosa depth were measured using a micrometer and recorded by a pathologist. The pathologist was blinded to the surgical technique used and final diagnosis, however, could not be blinded to the absolute size and depth of the specimen. Percent submucosa ([submucosa depth/total depth] × 100%) was calculated to determine whether a specimen would be labeled as adequate or inadequate by published pathology standards [16–18]. For this study, specimens were labeled adequate if they were found to have ≥50% submucosa [18]. Specimens not meeting this criterion were labeled inadequate. Volume was calculated from gross measurements to determine relative sizes of suction and full-thickness specimens. Diagnosis was verified as positive for HD only after a minimum of 100 serial sections