Massive bleeding after rectal suction biopsy: uncommon and unexpected delayed onset

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Background: **Rectal suction biopsy is a safe and painless procedure commonly performed in pediatric surgical practice for the diagnosis of intestinal dysganglionosis.**

Methods: **We report a 3.5-year-old boy who experienced massive delayed bleeding after a rectal suction biopsy. Detailed information regarding clinical features, onset, management, and outcome are provided.**

Results: **Acute onset of massive bleeding occurred 4 days after a rectal suction biopsy was performed to rule out possible intestinal dysganglionosis. The complication was managed conservatively, but blood transfusion and hospitalization were required. No predisposing abnormalities were detected.**

Conclusions: **Massive bleeding after rectal suction biopsy is a well-known life-threatening complication. Nonetheless, such a delayed bleeding (up to 4 days after a rectal suction biopsy) has never been reported before. Parents should be made aware of this possibility and surgeons should be aware of this complication which can be scarcely predicted based on patient's and familial history.**

World J Pediatr 2011;7(1):83-85

Key words: **complications;**
diagnosis;
Hirschsprung;
intestinal dysganglionosis;
rectal suction biopsy

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doi:10.1007/s12519-011-0251-2

Introduction

Rectal suction biopsy (RSB) is widely employed in the diagnosis of intestinal dysganglionosis.[1-3] It represents a painless, safe, and effective technique that can be accomplished at the bed-space or in the outpatient setting without anesthesia or sedation.[4,5] Complications are rare and include specimen inadequacy, rectal perforation, and bleeding.[5,6] The latter is experienced by less than 1% of patients undergoing RSBs and usually occurs immediately after the procedure.[2,4,5-8] Rarely, bleeding persists and requires blood transfusion or some sort of intervention (diathermy or stitching of the bleeding wound).[2] We report a patient who experienced severe delayed rectal bleeding several days after a RSB. To our knowledge, such a delayed bleeding onset has never been reported. We thus provide detailed clinical information and discuss clinical implications of this uncommon occurrence.

Case report

We presented a delightful 3.5-year-old boy who was born at term by uncomplicated vaginal delivery. His parents were unrelated, and there was no remarkable familial history. He passed meconium within the first 24 hours of life. Shortly after birth he began complaining difficulties in passing stool and worsening constipation. He underwent several trials of ineffective pharmacological treatment with laxatives, enemas, and diet. According to the previously published criteria[1] he was scheduled for a RSB to be performed awake without sedation in the outpatient setting. No family history of bleeding disorder was described. Similarly, the child's past medical history was unremarkable without previous episodes of unexplained or excessive bleeding.

He therefore underwent two RSBs at 3 and 5 cm from the anal verge with the instrument Solo-RBT, according to the methodology previously published.[4] Immediately after the procedure the child experienced slight transient self-limited rectal bleeding that settled spontaneously. He was subsequently discharged without symptoms 6 hours after the RSB.

The patient remained asymptomatic for the following three days. His parents described a couple of slightly blood stained bowel movements. Four days after the procedure he presented to the Accident and Emergency Department of our hospital due to the recent acute onset of massive rectal bleeding with incoming hypovolemic shock. At admission he was pale, hypotonic, tachycardic, hypotensive, with poor peripheral perfusion. He was resuscitated with administration of crystalloids (20 ml/kg bolus of saline) and 15 ml/kg of red blood cells (hemoglobin dropped to 7.4 g/dl before transfusion). He was also administered intravenous tranexamic acid (antifibrinolitic agent) to control bleeding. An abdominal ultrasound scan and X-ray performed at admission ruled out gross anatomical abnormalities, free fluid, free air, and portal hypertension. Fortunately, rectal bleeding settled and the child recovered quickly without further occurrences of bleeding and without the need for any surgical intervention to control bleeding. Clotting capacity (including International Normalised Ratio, activated partial thromboplastin time, and fibrinogen) determined at admission was within normal ranges. He was therefore discharged after 3 days of hospitalization in good general conditions, with normal bowel movements, asymptomatic.

Two months later, the child underwent a further elective screening for clotting capacity, which only detected partial deficiency of Factor XII (51% of total activity) and normal overall platelet function.

The histological assessment of the specimen excluded the presence of vascular abnormalities or chronic inflammation within the harvested specimen (Fig. 1). Histochemical staining showed unspecific innervative abnormalities which did not completely fulfil the criteria for any known intestinal dysganglionosis (pathological finding: isolated increased acetylcholinesterase positive fibers in the lamina propria and around submucosal vessels without hypertrophic submucous plexus, giant ganglia, or heterotopic ganglia) (Fig. 2). The child was therefore started on conservative treatment with oral laxatives (polietilenglicole 4000 at a dosage of 1 g/kg once daily) and cleansing phosphate enemas with increasing improvement.

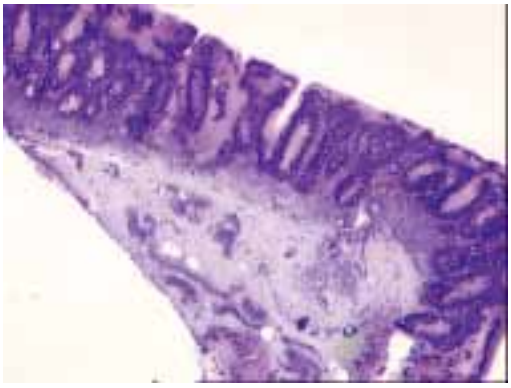

Fig. 1. Toluidine blue section showing an adeguate size rectal suction biopsy without enlarged vessels. Similarly, no infiltrate can be seen in the slide, thus excluding chronic rectal inflammation to justify a massive delayed bleeding (original magnification × 25).

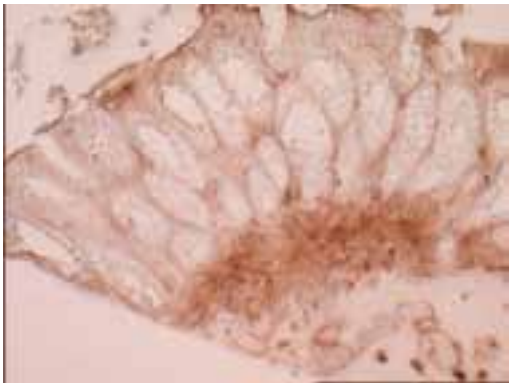

Fig. 2. Acetylcholinesterase section showing increased activity in the lamina propria and muscularis mucosae, some fibers around submucosal vessels but no hypertrophic submucous plexus. A small ganglion is evident underneath the muscularis mucosae (original magnification × 40).

Discussion

Rectal bleeding is a well-known complication of RSB. Its incidence varies but remains lower than 1% according to different series.[2,4,5-8] It has been described during or immediately after the procedure to require either blood transfusion, diathermy, packing, stitching, or nothing at all. Such a delayed rectal bleeding has never been described in previous reports.

It is well-known that every surgical procedure can be complicated by bleeding that, in some instances, can represent the onset of underlying clotting defects. In the present case the laboratory screening turned out to be poorly suggestive for any known clotting disorder. In fact, the slight deficiency of factor XII (Hageman factor), per se, should not predispose to abnormal or delayed bleeding.[9] Similarly, other predisposing causes of delayed/abnormal bleeding such as portal hypertension, full-thickness biopsy, or abnormal rectal vasculature (hemangioma or chronic inflammation)[4] can complicate any major or minor surgical procedure. In the present case these causes have been ruled out with adequate investigations. Therefore, this complication should be directly related to the procedure and represents a sentinel event alert for a common procedure in pediatric surgical practice.

In our experience, this was the first delayed bleeding in over more than 600 patients. Although

many reports described rectal bleeding as a possible complication of RSB, none underlined the possibility of a delayed onset of this complication, whose incidence seems not to be interfered by the instrument employed to perform the procedure.[2,4,5-8] In fact, immediate and/or prolonged bleeding occurred in 0.5% of patients from the series (using the instrument Solo-RBT) we previously published.[4]

We think that parents of patients undergoing a rectal suction biopsy should be aknowledged regarding the possibility of a delayed bleeding which would presumably occur at home. Either immediate or delayed, bleeding usually settles spontaneously and only requires supportive treatment. Rarely, packing of the rectum with gauzes moistened in antifibrinolitic agents, stitching, or diathermy can be necessary to control bleeding.

In our opinion RSB remains a safe, painless, and reliable procedure that should be accomplished in an outpatient setting without the need for any preoperative testing nor anaesthesia, provided personal or familial history is unremarkable.[4] Due to the extreme rarity of this complication, we did not modify our protocol for RSB. Similarly, we do not think that anoscopy or other diagnostic procedures can be useful to rule out bleeding that usually presents spontaneously due to the well-known irritative effect of blood on rectal mucosa. We simply suggest to maintain the patient in the ward or in the outpatient setting for 1 to 2 hours after the procedure unless signs of complications occur and to discharge the patient, when asymptomatic. Newborns are admitted for longer periods due to the likelihood of increased risk of complications.

Nonetheless, this occurrence prompted us to provide every family of patients undergoing RSB with a brief discharge summary describing any possible complication including delayed bleeding. Based on the previously published,[4] it is of utmost importance to reassure the parents and any physician dealing with the child of the usually self-resolving feature of this complication that rarely requires aggressive treatment.

In conclusion, delayed bleeding after a rectal suction biopsy can be a really severe and life-threatening complication. Parents or patients should be aware of this, though bleeding tends to settle spontaneously with the child only requiring supportive treatment.

Funding: None.
Ethical approval: Not needed.
Competing interest: None declared.
Contributors: Pini-Prato A wrote the main body of the article under the supervision of Carlini C. Pesce F and Jasonni V provided advice on medical aspects. Seymandi P is the guarantor.

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Received May 18, 2009
Accepted after revision July 27, 2009