# Rectal red blood loss in a healthy toddler is not always a juvenile polyp

T. Rousseff[1] , L. Matthyssens[2] , K. Van Renterghem[2] , D. Van de Putte[2] , D. De Wulf[3] , M. Van Winckel[1] , S. Van de Velde[1], R. De Bruyne[1] , S. Van Biervliet[1]

Departments of (1) Pediatric Gastroenterology and (2) Pediatric Surgery/GIHK, Princess Elisabeth Children's Hospital, Ghent, Belgium ; (3) Endoscopic Unit, Ghent University Hospital, Ghent, Belgium.

## Abstract

**Aim Heterotopic gastric mucosa is a well-known congenital anomaly in Meckel's diverticula and duplication cysts. Solitary heterotopic gastric mucosa in the rectum is a rare and frequently overlooked abnormality. Starting from a patient history, the literature is searched and all cases reported over the past 20 years are reviewed and compared to a summary of the older cases. Differences between adult and childhood presentation are outlined and our patient is compared with prior reported cases. Case A 3-year-old girl presented with recurrent rectal blood loss caused by heterotopic gastric mucosa without duplication cyst. She was endoscopically treated with two-stage endoscopic surgical dissection (ESD). Up to now, rectal heterotopic gastric mucosa has been reported in 34 adults and 24 children, including this patient. There is an overall male dominance (69%). Presenting complaints in children were recurrent fresh blood loss per anum (96%), pain (46%), perineal ulcers (25%), diarrhoea (8%) and one patient had an ano-cutaneous fistula. Endoscopy revealed a mucosal elevation with a slightly different aspect (33%), a polyp (42%) and a solitary ulcer (25%). Endoscopy in adults reveals more frequently polyps compared to children. Treatment in childhood is mainly surgical where adults are more frequently treated with endoscopic techniques. Conclusion In a child with recurrent rectal bleeding in good general health, it is important to withhold heterotopic gastric mucosa in the differential diagnosis and take sufficient biopsies during endoscopy.** (Acta gastroenterol. belg., **2017**, 80, **67-70**).

**Key words**: heterotopic gastric mucosa, rectal blood loss, children

## Introduction

Small amounts of faecal fresh red blood loss, in an otherwise healthy child without constipation, suggest the presence of a juvenile rectal polyp. Other causes such as colitis, vascular abnormalities and fissures are often associated with other symptoms. Recto-sigmoidoscopy is indicated for diagnosis and eventual simultaneous treatment. The following case points out the importance of taking sufficient biopsies, even in case of minor endoscopic abnormalities. An overview of all recently reported cases of rectal heterotopic gastric mucosa is given.

## Case report

A 3-year-old girl in a good general health consulted with recurrent faecal red blood loss since one year. The blood was mixed with mucous stools. She complained intermittently of tenesmus. Defaecation was regular and tenesmus without other complaints. Anal inspection as well as digital rectal exam were normal. A stool sample was negative for enteropathogens. Rectosigmoidoscopy could not detect a polyp but revealed a well-defined aberrant mucosa 1.5 cm from the margo ani (Figure 1 and 2) of which biopsies were taken. Pathologic examination revealed ectopic gastric mucosa.

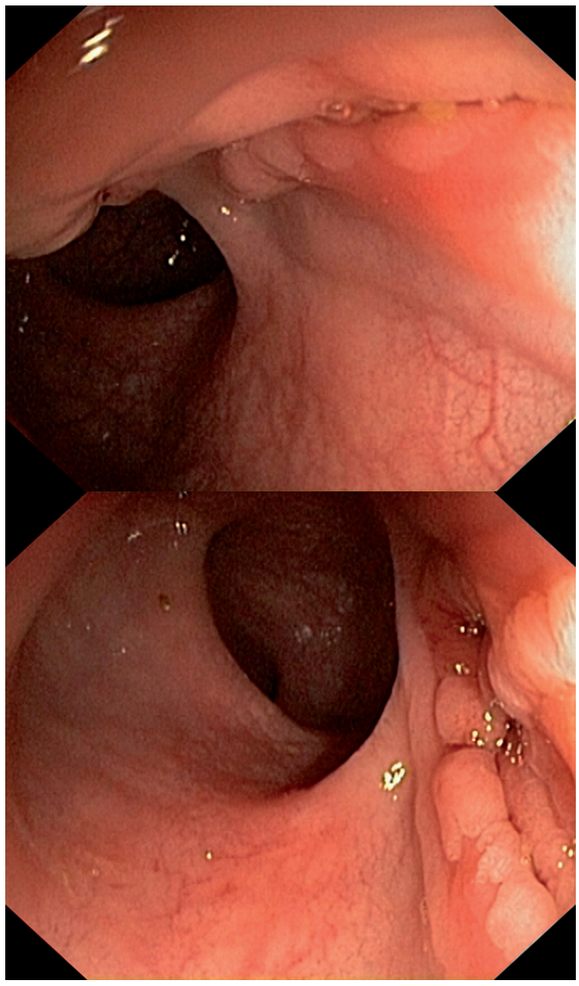

Fig. 1 and 2 — Well-defined lesion of aberrant mucosa (3 x 2 cm) at the right lateral rectal wall, 1.5 cm from the margo ani.

Correspondence to : Stephanie Van Biervliet, UZ Gent, De Pintelaan 185, 9000 Gent, Belgium
E-mail : Stephanie.vanbiervliet@ugent.be

Submission date : 18/01/2016
Acceptance date : 15/10/2016

Endoscopic treatment with argonplasma coagulation was unsuccessful, whereupon omeprazole was started to diminish the local erosive effects. After an asymptomatic period of 10 months, the same symptoms recurred despite omeprazole. An endoscopic submucosal dissection (ESD), lifting the mucosa with saline 0,9% was performed. Histologic examination showed colonic mucosa associated with gastric corpus-fundic type mucosa. Following this procedure she has remained asymptomatic.

## Discussion

Heterotopic gastric mucosa is a congenital anomaly and a well-known phenomenon in Meckel's diverticula or duplication cysts. Since the first paediatric report of heterotopic gastric mucosa in the rectum in 1939 by Ewell et al (1), only 24 paediatric cases are described (1-9, 12). The number of reported cases is rapidly increasing since the availability of endoscopy. As interventional endoscopy has known an important evolution over the past 20 years, the cases reported since 1994 (9 children and 23 adults) were reviewed and compared to prior reports (10,14). Results are given in Table 1 (children) and Table 2 (adults).

There is a male predominance (childhood 70% , adults 68%). The clinical presentation differs according to age. All children were symptomatic presenting with fresh rectal blood loss (2-9). This complaint was eventually associated with diarrhoea (22%), tenesmus (22%) and abdominal pain (55%). In adults one in three is asymptomatic and 16% has occult blood loss. Adults complain most frequently of pain (39%), blood loss (35%) and diarrhoea (13%). Symptomatology did not change over time. Median age at diagnosis in children decreased from 7 to 4 years, reflecting the availability of endoscopy. As a result, the delay between onset of symptoms and diagnosis also decreased.

The endoscopic lesions in children were aberrant mucosa (45%), a solitary ulcer (33%) and polyps (22%). In contrast the historical paediatric cases had polyps in 53%, an abnormal mucosa in 27% and ulcers in 20%. Adults displayed polyps (65%) and aberrant mucosa (35%). The change in paediatric endoscopic presentation over time as well as the adult presentation could point to an evolution of the lesion over time. However, improved endoscopic quality, detecting more subtle mucosal changes cannot be excluded.

Table 1. — **Overview of recently reported (since 1994) paediatric cases of rectal heterotopic gastric mucosa as well as a summary of the older cases**

| **Reference (publ. year)** | **Gender, age** | **Symptoms** | **Endoscopic Finding** | **Histopathology** | **Treatment** |
|---|---|---|---|---|---|
| (2) (2002, Wiersma) | Boy, 2 years | RBPA | Shallow pit Ulcer | Type not specified | Surgery |
| (3) (2004, De Angelis) | Boy, 4,5 years | RBPA Pain Diarrhoea | Ulcer | Mixed type mucosa | Medication Endoscopic mucosectomy |
| (4) (2007, Garmendia) | Boy, 4 years | RBPA Pain Diarrhoea | Elevated aberrant mucosa | Type not specified | Medication Surgery |
| (5) (2007, Cheli) | Girl, 2 years | RBPA Pain | Elevated aberrant mucosa | Type not specified | Medication Surgery |
| (6) (2010, Sauer) | Girl, 5 years | RBPA | Polyp | Fundic type mucosa | Endoscopic ablation Medication Endoscopic mucosectomy |
| (7) (2010, Di Nardo) | Boy, 6 years | RBPA Pain Tenesmus | Elevated aberrant mucosa | Type not specified | Medication Surgery |
| (8) (2011, Kokil) | Boy, 12 years | RBPA | Polyp | Fundic type mucosa | Surgery |
| (9) (2014, Al-Hussaini) | Boy, 3 years | RBPA Pain | Ulcer | Fundic type mucosa | Surgery |
| Current Case | Girl, 3years | RBPA Tenesmus | Elevated aberrant mucosa | Mixed type mucosa | Endoscopic ablation Medication Endoscopic mucosectomy |
| **Summary** 9 paediatric cases | 4,6 years (2y-12y) 6 M, 3 F | 9 RBPA 5 Pain 2 Diarrhoea 2 tenesmus | 4 Aberrant mucosa 3 Ulcers 2 Polyp | 3 Fundic 2 Mixed 4 Not specified | 6 Surgery 3 Endoscopic |
| **Literature** 15 paediatric cases before 1994 (10, 14) | 7 years (1d-17 y) 11 M, 4 F | 14 RBPA 6 Pain 6 Perineal ulcer 1 Fistula | 8 Polyp 4 Aberrant mucosa 3 Ulcer | 10 Fundic type 1 Corpus type 1 Mixed type 3 Unspecified | 13 Surgery 1 Trans-anal ablation 1 Medication |

RBPA: red blood loss per anum

Table 2. — **Overview of recently reported (since 1994) adult cases of rectal heterotopic gastric mucosa as well as a summary of the old cases**

| **Reference (publ. year)** | **Gender, age** | **Symptoms** | **Endoscopic Finding** | **Histopathology** | **Treatment** |
|---|---|---|---|---|---|
| (10) (1994, Devereaux) | Male, 31 years | Pain Diarrhoea | Elevated aberrant mucosa | Corpus type mucosa | Medication |
| (11) (1994, Srinivasan) | Male, 34 years | Pain | Depressed aberrant mucosa | Fundic type mucosa | Surgery |
| (12) (1999, Campo) | Female, 36 years | RBPA Pain | Polyp | Mixed corpus-fundic type mucosa | Endoscopic loop resection |
| | Male, 34 years | Anaemia | Polyp | Fundic type mucosa | Endoscopic loop resection |
| (13) (2000, Lascar) | Male, 48 years | RBPA | Polyp | Fundic type mucosa | Endoscopic mucosectomy |
| (14) (2004, Steele) | Male, 21 years | Pain | Polyp | Fundic type mucosa | Medication Surgery |
| (15) (200, Vieth) | Male, 46 years | Asymptomatic | Polyp | Corpus type mucosa | Endoscopic loop resection |
| (16) (2006, Davidoff) | Male, 60 years | Asymptomatic | Polyp | Mixed corpus-fundic type mucosa | Medication |
| (17) (2007, Ikematsu) | Male, 69 years | Occult blood | Depressed aberrant mucosa | Fundic type mucosa | Endoscopic mucosectomy |
| (18) (2009, Corrigan) | Female, 47 years | RBPA Diarrhoea | Elevated aberrant mucosa | Mixed corpus-fundic type mucosa | Medication Endoscopic ablation |
| (19) (2010, Limdi) | Female, 36 years | Pain | Polyp | Type not specified | Endoscopic resection |
| (20) (2011, Reis) | Male, 53 years | Asymptomatic | Elevated aberrant mucosa | Corpus type mucosa | Endoscopic mucosectomy |
| (21) (2011, Marin) | Male, 22 years | RBPA | Polyp | Corpus type mucosa | Surgery |
| (22) (2011, Yamagishi) | Female, 68 years | Occult blood | Flat aberrant mucosa | Fundic type mucosa | Endoscopic mucosectomy |
| (23) (2012, Jotautas) | Male, 57 years | RBPA Pain | Polyp | Type not specified | Surgery |
| (24) (2012, Cheng) | Male, 57 years | RBPA Pain | Polyp Ulcer | Type not specified | Surgery |
| (25) (2012, Kim) | Male, 19 years | RBPA Pain | Polyp Ulcer | Mixed corpus - fundic type mucosa | Surgery |
| (26) (2012, Huelsen) | Male, 46 years | Asymptomatic | Flat aberrant mucosa | Corpus type mucosa | None |
| | Male, 51 years | Diarrhoea | Polyp | Corpus type mucosa | None |
| (27) (2012, Assimakopoulos) | Male, 48 years | Pain | Polyp | Fundic type mucosa | Endoscopic mucosectomy |
| (28) (2015, Swatec) | Female 58 years | Asymptomatic | Polyp | Fundic type mucosa | Endoscopic loop resection |
| (29) (2015, Chrysanthos) | Male 22 years | RBPA | Polyp | Corpus Type | Surveillance, medication |
| (30) (2016, Iacopini) | Male 63 years | Asymptomatic | Aberrant mucosa | Corpus Type | Endoscopic mucosectomy |
| **Summary** 24 adult cases | 47 years (19-69y) 18 M, 5 F | 8 Asymptomatic 9 Pain 8 Blood loss 3 Diarrhoea | 15 Polyp 8 Aberrant mucosa 2 Ulcer | 8 Fundic type 8 Corpus type 4 Mixed type 3 Unspecified | 12 Endoscopic 6 Surgery 3 Medication 3 No treatment |
| **Literature** 11 adult cases published before 1994 (10,14) | 25 years (19-51y) 5 M, 6 F | 3 Asymptomatic 8 Blood loss 4 Pain | 5 Polyp 3 Aberrant mucosa 3 Ulcer | 5 Fundic type 2 Corpus type 3 Mixed type 1 Pyloric type | 9 Surgery 1 Endoscopy 1 Medication |

RBPA: red blood loss per anum

Although some historical cases report multiple gastrointestinal locations of heterotopic gastric mucosa (31, 32), a $^{99m}$Tc scan was reported in only 9/27 recent patients. Seven confirmed the isolated heterotopic gastric mucosa in the rectum (3, 7, 11, 14, 19, 21, 23), one revealed a second endoscopic unconfirmed locus of $^{99m}$Tc uptake (11) and 2 were false negative (2, 4). Given these numbers, one could question whether it is necessary to submit all patients to this test.

Histology in adults revealed fundic type mucosa (3 %), corpus type mucosa (35%) and mixed mucosa (17%). In paediatric cases, only fundic type mucosa (60%) and mixed mucosa (40%) were described.

Removing the heterotopic gastric mucosa was the treatment of choice in children as they were all symptomatic. The majority was treated by transanal surgery (67%), only 3 patients (33%) were treated endoscopically. In adults treatment was more diverse. Endoscopic removal was performed in 52%, surgery in 26%, medication was given in 9% and no treatment in 13%. The observed difference in treatment modality between adult and paediatric cases is probably due to less acquaintance with endoscopic mucosectomy in paediatrics as indications for this technique are scarce. The therapy in adults has made an important shift from surgery (82%) in the older literature towards endoscopic removal recently. Despite the description of focal intestinal metaplasia (25) as well as differentiated carcinoma (15) in adults with rectal heterotopic gastric mucosa, there remains some discussion whether the incidental finding of asymptomatic heterotopic gastric mucosa in the rectum should be treated or just followed (25). As children diagnosed with heterotopic gastric mucosa in the rectum are always symptomatic and most patients relapse during acid suppressive therapy, resection of the heterotopic mucosa is considered the treatment of choice (2). The shift towards endoscopic resection of the mucosa in children has started recently with only 3 reports on endoscopically treated children up to now (3, 7, current case).

## Conclusion

Heterotopic gastric mucosa in the rectum is a known but rare cause of rectal blood loss in children. Colonoscopic investigation with sufficient biopsies even in presence of minimal mucosal changes is necessary to make the diagnosis. Resection of the heterotopic mucosa resolves symptoms.

## References


1. EWELL G.H., JACKSON R.H. Aberrant gastric mucosa in the rectum with ulceration and hemorrhage. *Wisc. Med. J.*, 1939, **38** : 641-3.
2. WIERSMA R., HADLEY G.P., GOVENDER D., GRANT H.W. Rectal gastric heterotopia in infancy. *J. Pediatr. Surg.*, 2002, **37** : 1481-2.
3. DE ANGELIS P., TRECCA A., FRANCALANCI P., TORRONI F., FEDERICI DI ABRIOLA G., PAPADATOU B., CIOFETTA G.C., DALL'OGLIO L. Heterotopic gastric mucosa of the rectum. *Endoscopy*, 2004, **36** : 927.
4. GARMENDIA M., APARICO J.R., MARTINEZ J., CASELLAS J.A. *Gastrointest. endos.*, 2007, **65** : 921-2.
5. CHELI M., ALBERTI D., VAVASSORI D., SONZOGNI A., PEDRETTI E., LOCATELLI G. Heterotopic rectal gastric mucosa: a rare cause of lower gastrointestinal bleeding in children. Case report and review of pediatric literature. *Eur. J. Pediatr. Surg.*, 2007, **17** : 50-4.
6. SAUER C.G., BICKSTON S.J., BOROWITZ S.M. Gastric Heterotopia of the rectum. *J. Ped. Gastroenterol. Nutr.*, 2010, **50** : 329-33.
7. DI NARDO G., FREDANI S., LUCARELLI S., COZZI D., OLIVA S., MARCHEGGIANO A., CUCCHIARA S. An unusual cause of rectal bleeding in a child. *Gastrointest. Endosc.*, 2010, **72** : 851-2.
8. KOKIL G., PULIMOOD A., MATHAI J. A rare case of a rectal polyp with gastric heterotopia. *Indian J. Pathol. Microbiol.*, 2011, **54** : 848-9.
9. AL-HUSSAINI A., LONE K., AL-SOFYANI M., EL BAGIR A. Gastric heterotopia of rectum in a child : a mimicker of solitary rectal ulcer syndrome. *Ann. Saudi Med.*, 2014, **34** : 245-9.
10. DEVEREAUX C.E., DEVEREAUX R.G. Heterotopic gastric mucosa of the rectum with a review of the literature. *J. Clin. Gastroenterol.*, 1994, **19** : 41-5.
11. CAMPO R., BRULLET E., MONTANE J.M., PANADES A. Uncommon appearance of gastric mucosal ectopia in the rectum. *Gastrointest. Endosc.*, 1994, **40** : 653-4.
12. SRINIVASAN R., LOEWENSTINE H., MAYLE J.E. Sessile polypoid gastric heterotopia of rectum: a report of 2 cases and review of the lieterature. *Arch. Path. Lab. Med.*, 1999, **123** : 222-4.
13. LASCAR G., HOUISSA-VUONG S., MARTIN B., VUONG P.N. Hétérotopie gastrique dans le rectum: un nouveau cas. *Gastroenterol. Clin. Biol.*, 2000, **24** : 849-50.
14. STEELE S.R., MULLENIX P.S., MARTIN M.J., ORMSETH E., WEPPLER E., GRAHAM J., PLACE R.J. Heterotopic gastric mucosa of the anus: a case report and review of the literature. *Am. Surgeon*, 2004, **70** : 715-9.
15. VIETH M., KUSHIMA R., DE JONGE J.P.A., BORCHARD F., OELLIG F., STOLTE M. Adenoma with gastric differentiation (so-called gland adenoma) in a heterotopic gastric corpus mucosa in the rectum. *Virchows Arch.*, 2005, **446** : 542-5.
16. DAVIDOFF S, JALAL PK, GAO Z, KOPS RJ, ROBINS T, BANK S. Gastric heterotopia in the rectum. *Gastrointestinal endoscopy*, 2006, **63** : 1075.
17. IKEMATSU H., FU KI, SAITO Y., MATSUDA T., SHIMODA T., FUJII T. Ectopic gastric mucosa in the rectum mimicking an early depressed cancer treated by endoscopic mucosal resection. *Endoscopy*, 2007, **39** : E171-2.
18. CORRIGAN MA, SHIELDS CJ, KEOHANE C, KIRWAN WO. The immunohistochemical demonstration of Helicobacter pylori in rectal ectopia. *Surg. Laparosc. Endosc. Percutan. Tech.*, 2009, **19** : e146-8.
19. LIMDI J.K., SAPUNDZIESKI M., CHAKRAVARTHY R., GEORGE R. Gatsric heterotopia in the rectum. *Gastrointest. Endosc.*, 2010, **72** : 190-1.
20. REIS H., KERKER J., SCHAEFER A., BUDDE T., SCHMID K.W., METZ K. Rektale magenschleimhautheterotopie: ein fallbericht in zusammenschau mit der literature. *Pathologe*, 2011, **32** : 521-4.
21. MARIN R.M., CANDEL ARENAS M.F., PARRA BANOS P.A., GONZALES VALVERDE F.M., RODENAS MONCADA J., BENAVIDES BULEJE J.A., MARTINEZ CRESPO J.J., QUIRANTE F.P., MARIN BLAZQUEZ A.A. Gastric heterotopia in the rectum: a rare cause of rectal bleeding. *Am. Surg.*, 2011, **77** : 659-62.
22. YAMAGISHI H., FUKUI H., TOMITA S., ICHIKAWA K., IMURA J., ISHIZUKA M., KUBOTA K., FUJIMORI T. *Intern. Med.*, 2011, **50** : 1587-9.
23. JOTAUTAS V., RUTKAUSKAITÉ D., STRUPAS K. Transanal endoscopic microsurgery – a new method in the treatment of complicated large gastric heterotopy in the rectum. *Int. J. Colorectal. Dis.*, 2012, **27** : 1539-40.
24. CHENG D.W., SEKHON H.K., TOUTOUNJIAN R., ABBAS M.A. Heterotopic gastric mucosa of the anus associated with anal ulcer. *Tech. Coloproctol.*, 2012, **16** :167-8.
25. KIM H.S., KIM J.H., YOON S.O. Heterotopic gastric mucosa with focal intestinal metaplasia and squamous epithelium in the rectum. **Dig. endosc.**, 2012, **24** : 46-8.
26. HUELSEN A., FALVEY J., WHITEHEAD M., DING S. Gastrointestinal : large heterotopic gastric mucosa in the rectum. *J. Gastroenterol. Hepatol.* 2012, **27** : 1641.
27. ASSIMAKOPOULOS S.F., KOUREA H.P., GOGOS C.A., THOMOPOULOS K.C. Gastrointestinal : a rare cause of anal pain: gastric heterotopia of the rectum. *J. Gastroenterol. Hepatol.*, 2013, **28** : 1432.
28. SWATEC J., WRONECKI L., CIECHANEK R., SZUMILO J. Asymptomatic gastric heterotopias in the rectum with Helicobacter Pylori infection. *Pol. J. Pathol.*, 2015, **66** : 426-9.
29. CHRYSATHOS N,. ANAGNOSTOPOULOU E., DASKALAKI A., PATSAVELA S. Heterotopic gastric mucosa of the rectum presenting as rectal bleeding. *Am. J. Gastroenterol.*, 2015, **110** : 498.
30. IACOPINI F., GOTODA T., ELISEI W., RIGATO P., MONTAGNESE F., SAITO Y., COSTAMAGNA G., IACOPINI G. Heterotopic gastric mucosa in the anus and rectum : first case report of endoscopic submucosal dissection and systematic review. *Gastroenterol. Rep.* (Oxf), 2016 Apr 21. pii: gow006.
31. PICARD E.J., PICARD J.J., JORISSEN J., JARDON M. Heterotopic gastric mucosa in the epiglottis and rectum. *Am. J. Dig. Dis.*, 1978, **23** : 217-21.
32. MURRAY F.E., LOMBARD M., DERVAN P., FITZGERALD R.J., CROWE J. Bleeding from multifocal heterotopic gastric mucosa in the colon controlled by an H2 anatagonist. *Gut*, 1988, **29** : 848-51.