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Digestive perianastomotic ulcerations and Crohn's disease



Marie-Louise Frémond^a, Jérome Viala^{a,b,c,1}, Xavier Tréton^{b,c,d,1}, Maryline Roy^{b,c}, Dominique Berrebi^{a,b,c}, Frédéric Gottrand^e, Arnaud Bonnard^{a,b}, Christine Martinez-Vinson^{a,1}, Jean-Pierre Hugot^{a,b,c,*,1}

Received 18 February 2014; received in revised form 29 June 2014; accepted 30 June 2014

KEYWORDS

Digestive perianastomotic ulcerations; Crohn's disease; Ulcers; Ileocolonic anastomosis; Small bowel surgery; Nod?

Abstract

Background and aims: Digestive perianastomotic ulcerations (DPAU) have been occasionally reported as late complications of neonatal or childhood surgery.

Methods: We report here a series of 14 new cases.

Results: Cases were revealed by severe anemia, diarrhea, abdominal pain and growth failure in average 11.5 years after surgery. Ulcerations were most often multiple (n = 11), located on the upper part of ileocolonic anastomoses (n = 12) and difficult to treat. No granulomas were seen but lymphoid follicles were frequent. In addition, either ASCA or ANCA were positive in 4/9 tested patients and 8/11 genotyped patients exhibited a NOD2 mutation (P < 0.0002 when compared to French healthy controls).

Conclusion: Altogether, these findings argue for common physiopathological features between DPAU and Crohn's disease and for a prospective follow-up of selected operated children to explore the early events involved in gut inflammatory lesions.

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^a Assistance Publique—Hôpitaux de Paris, Services des Maladies Digestives et Respiratoires de l'enfant, Service de Chirurgie Digestive et Service de Cytologie et Pathologie, Hôpital Robert Debré, 75019 Paris, France

^b Université Paris-Diderot UMR 1149, Labex Inflamex, 75018 Paris, France

^c INSERM UMR1149, Centre de Recherche sur l'inflammation Paris Montmartre, 75018 Paris, France

^d Assistance Publique—Hôpitaux de Paris, Service de Gastroentérologie, Hôpital Beaujon, 92000 Clichy, France

^e Inserm U995, Hôpital Jeanne de Flandre, 59000 Lille, France

^{*} Corresponding author at: Service des maladies digestives et respiratoires de l'enfant, Hôpital Robert Debré, 48 Bd Sérurier, 75019 Paris, France. Tel.: +33 1 40 03 57 12; fax: +33 1 40 03 57 66.

E-mail address: jean-pierre.hugot@rdb.aphp.fr (J.-P. Hugot).

¹ These authors equally contributed to the work.

1. Introduction

Digestive perianastomotic ulcerations (DPAU) are late complications which occur after neonatal or childhood intestinal surgery. They are rare events out of the context of inflammatory bowel disease (IBD) and more specifically Crohn's disease (CD). The first cases were described by Parashar et al. in 1988¹. We performed a literature review by the interrogation of the Pubmed database, using the

following key words: ileocolonic/perianastomotic/postsurgical ulcerations/ulcers. Thirteen papers reporting a total of 27 cases were retrieved and analyzed $^{1-13}$.

To further document this rare condition, we reviewed a series of 14 cases followed at hospitals Jeanne de Flandre (Lille, France) Robert Debré and Beaujon (Paris, France). Clinical, biological, radiological, endoscopic and histological features were retrospectively collected from patients' medical files. This work complied with the Declaration of Helsinki and patients provided an informed consent for genetic studies.

Table 1 Summary of the reported PDAU cases. 5-ASA: 5-aminosalicylic acid; AB: antibiotherapy; ASCA: anti-saccharomyces cerevisiae antibodies; ANCA: antineutrophil cytoplasmic antibodies; AZA: azathioprine; BT: blood transfusion; C+: colonic ulceration; CIPO: chronic intestinal pseudo-obstruction; CRP: C-reactive protein; CS: corticosteroids; EN: enteral nutrition; ESR: erythrocyte sedimentation rate; F: treatment failure; GI: gastrointestinal; I: iron supplements; IFX: Infliximab; MTX: methotrexate; NEC: necrotizing enterocolitis; ND: not done; PN: parenteral nutrition; R: resection of PDAU; S: treatment with at least partial response (success).

Case	1	2	3	4	5	6	7
Actual age (y), gender	14, F	16, M	9, M	12, M	8, F	13, M	27, F
Age at onset (y)	11	12	5	11	6,5	6	26
Underlying disease	NEC	NEC	NEC	Hirschsprung	NEC	Hirschsprung	NEC
Ileocecal valve	No	No	No	No	No	Yes	No
Clinical features							
Diarrhea/GI bleeding	Yes/no	Yes/yes	Yes/no	Yes/yes	Yes/yes	Yes/yes	Yes/no
Abdominal pain/Koenig	Yes/yes	Yes/no	Yes/no	Yes/no	Yes/no	Yes/no	No/yes
Growth retardation	Yes	Yes	Yes	Yes	Yes	No	Yes
Perineal lesions	No	No	No	No	No	No	No
Documented stenosis	No	No	No	No	No	No	No
Biology and genetics							
Hemoglobin (g/l)albuminemia (g/l)	95/27	80/38	93/21	59/16	61/23	115/48	129/32
CRP (mg/ml)ESR (mm)	48/50	5/15	22/26	178/26	<10/8	ND	6/24
ASCA/ANCA	+/-	-/-	-/-	-/-	-/+	ND/ND	ND/ND
NOD2 genotype	1007 fs	G908R	No mutation	No mutation	R702W	1007 fs	R702W
Endoscopy and histology							
Ulceration features	Multiple,	Multiple,	Multiple,	Multiple,	Multiple,	Unique,	Multiple,
	deep, C+	deep	superficial, C+	deep, C+	deep	deep	superficial
Lymphoid f.	Yes	No	Yes	Yes	Yes	No	No
Eosinophils	Yes	Yes	Yes	Yes	Yes	Yes	Yes
Abnormal architecture	Yes	Yes	Yes	Yes	Yes	No	Yes
Neovessels	No	No	Yes	Yes	No	No	No
Evolution and treatments							
Support treatments	AB, EN, PN	1	AB, EN	AB, BT, EN, PN, BT	AB, BT	I	EN
5-ASA	Yes (F)	Yes (S)	No	No	Yes (F)	No	Yes (S)
Budesonide	No	Yes (S)	No	No	No	No	No
Systemic steroids	Yes (S)	No	Yes (S)	Yes (S)	Yes (F)	No	Yes (S)
Immunosup-pressors and biotherapies	AZA (F)	No	AZA (F)	No	AZA (F)	No	No
Surgery	No	No	Yes (ileo-stomy, S)	No	Yes (R, F)	Yes (S)	No
Follow-up (y)	3.5	3.5	3	1.3	1.6	4	1
Today remission	No	No	Yes	Yes	No	Yes	No

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