

CASE REPORTS

Inflammatory bowel disease in married couples: 10 cases in Nord Pas de Calais region of France and Liège county of Belgium

M C Comes, C Gower-Rousseau, J F Colombel, J Belaïche, H J Van Kruiningen, M C Nuttens, A Cortot

Registre des Maladies Inflammatoires du Tube Digestif du Nord Ouest de la France
CHRU Lille, France
M C Comes
C Gower-Rousseau
J F Colombel
M C Nuttens
A Cortot

Service d'Hépatogastroentérologie,
CHU Liège, Belgium
J Belaïche

Department of Pathobiology,
University of Connecticut, Storrs,
USA
H J Van Kruiningen

Correspondence to:
Professor J F Colombel,
Registre des Maladies Inflammatoires du Tube Digestif du Nord Ouest de la France, Service d'Hépatogastroentérologie, Hôpital Huriez, CHRU Lille, 59037 France.

Accepted for publication
8 December 1993

Abstract

Ten pairs of husband-wife couples are reported with inflammatory bowel disease who were seen in the same geographical area in Nord Pas de Calais region of France and in Liège county (Belgium). Among these 10 couples, four were concordant for Crohn's disease, two for ulcerative colitis, and four were discordant. In nine of 10 couples neither spouse had symptoms before marriage but inflammatory bowel disease subsequently developed in both. In one couple, one spouse had Crohn's disease before marriage and the other partner experienced symptoms afterwards. Eighteen children were born to eight of 10 couples. Five of them developed Crohn's disease but four belong to the same family. In all cases the affected children were born to parents who both developed Crohn's disease after they had married and were conceived at a time when parents did not yet have symptoms. It is proposed that this pattern of emergence of inflammatory bowel disease suggests a role for an infectious agent yet to be identified.

(Gut 1994; 35: 1316-1318)

Inflammatory bowel disease (IBD) has been ascribed to genetic influences, environmental factors, or a combination of both. The rarity of IBD in married couples has been considered evidence against the importance of environmental factors in the aetiology of the disease. In 1991, however, Bennett *et al* from New York identified 21 couples with IBD, 14 of whom experienced disease in the second spouse after they were married.¹ This study brought to 39 the total number of case reports of IBD in husband-wife couples in published works.²⁻¹⁸ We recently described the occurrence of Crohn's disease in a married couple, their four children and the wife of the eldest son.¹⁹ We now report an additional eight pairs of husband-wife couples with IBD seen in the same geographical area in Nord Pas de Calais region of France and in the county of Liège (Belgium).

Methods

This study was realised through the Registre des Maladies Inflammatoires du Tube Digestif du Nord-Ouest de la France²⁰ and the Liège county of Belgium. The total population of the geographical area (3 966 125 for Nord Pas de Calais and 441 125 for Liège County) was surveyed through the gastroenterologists. Each gastroenterologist participating in the study was asked to refer conjugal forms of IBD to an interviewer practitioner. All gastroenterologists were contacted - that is, 155, 147 of whom replied (95%). All the patients' family data were then collected in a standard form. The following data were gathered: dates of birth, marriage (or cohabitation), onset of symptoms, and diagnosis of IBD; consanguinity; location of residence; dates of birth of all children, type of IBD in parents and children. Diagnosis of IBD was established according to standard criteria.²⁰

Couples were assigned to three groups (A, B, C) paralleling the groups of Bennett *et al*¹ based on the time of onset of symptoms of disease in each partner relative to the data of the marriage: A, onset of symptoms occurred before marriage in both; B, onset of symptoms occurred in husband or wife after marriage; C, onset of symptoms occurred after marriage in both.

TABLE 1 Onset data for inflammatory bowel disease in spouses

	Sex	Year of birth	Onset of disease	Age at onset (y)	Interval from marriage to onset (y)	Type of IBD
B1*	M	1959	1974	15	NA	CD
	F	1961	1984	23	1	CD
C1*	M	1933	1970	37	12	CD
	F	1938	1988	50	30	CD
C2	M	1938	1980	42	21	CD
	F	1938	1990	52	31	CD
C3	M	1951	1992	41	18	CD
	F	1954	1990	36	16	CD
C4	M	1954	1991	37	7	CD
	F	1959	1985	26	1	UC
C5	M	1927	1981	54	23	CD
	F	1926	1992	66	44	UC
C6	M	1946	1983	37	10	UC
	F	1950	1975	25	2	CD
C7	M	1949	1988	39	9	CD
	F	1955	1980	25	1	UnC
C8	M	1947	1991	44	20	UC
	F	1951	1979	28	8	UC
C9	M	1943	1992	49	28	UC
	F	1942	1973	31	9	UC

* = Same family; UC = ulcerative colitis; CD = Crohn's disease; UnC = unclassified colitis; NA = not applicable.

Results

Eight couples who both had IBD were identified in Nord Pas de Calais and two in Liège County. None of the couples studied were consanguineous. No couple was assigned to group A, one couple, concordant for Crohn's disease, was assigned to group B, and nine to group C (Table I). In this group, three couples were concordant for Crohn's disease (C1, C2, C3), two for ulcerative colitis (C8, C9), and four were discordant (C4, C5, C6, C7). The mean age of disease onset was 30 years in the first spouse and 40 years in the second spouse. The first spouse to develop IBD was the husband in four couples (B1, C1, C2, C5) and the wife in the remaining six couples. The first spouse to develop IBD had Crohn's disease in six cases. The first spouse developed IBD an average of 9.4 years (1–23) after marriage and the second spouse an average of 10.4 years after the first.

Three of 20 husbands and wives with IBD had a first degree relative with IBD (wives of C1 and C4 couples and husband of B1 couple).

Eighteen children were born to eight of 10 couples (Table II). Among them five developed Crohn's disease, four belonging to the same family. In all cases the affected children were born to parents who both developed Crohn's disease after they had married and were conceived at a time when parents did not yet have symptoms of Crohn's disease.

Discussion

We report 10 married couples with IBD seen in the same geographical area of northern France and Liège county of Belgium. In nine of 10 couples neither spouse had symptoms before marriage, but IBD subsequently developed in both. In the last case only one spouse had IBD before marriage, and the other partner experienced symptoms afterwards. We found a high rate of IBD (28%) in the offspring of these couples, close to the 36% reported by Bennett *et al* but four of the five children belong to the same extensively described

family with remarkably similar Crohn's disease.¹⁹

Although improbable, we cannot confirm that these marital clusterings did not occur by chance. Using prevalence data of IBD in published works, Lobo *et al* and Bennett *et al* calculated that the occurrence of marital Crohn's disease in the United Kingdom and marital IBD in the United States was greater than expected by chance. No such calculation was possible in our study as the prevalence of IBD in France and Belgium is still unknown.

None of the couples were consanguineous and all were white thus avoiding the possible bias of unknown genetic links in our patients, as in Jews. Our series of marital IBD is thus certainly not suggestive of an inherited disease. On the contrary it may reflect an increase in environmental risk in the home. The mean interval between onset of disease in spouses was 10.4 years. A similar long latent period between new cases has been seen in other clusters of IBD. In one series of marital IBD the latent period was 6.4 years in group B and 6.8 years in group C.¹ In the other series, the latent period was 7.2 years (all couples belonging to group C).⁴ Clustering of cases of IBD among unmarried subjects has also been described. In 1986, Reilly and Robinson, from Northern Ireland, described four cases of Crohn's disease in unrelated adult women, ages 21–30, who had been close high school associates for seven years, from the ages of 11 to 18.²¹ Allan *et al* described a clustering of 12 patients with Crohn's disease in Gloucestershire, England.²² The latent period for Crohn's disease after leaving the community was three to five years. More recently Aisenberg *et al* described three unrelated men who enjoyed a close sustained friendship in college and who developed symptoms three, five, and nine years after the beginning of their contact.²³ We think that the pattern of emergence of IBD in our study and in these cases, suggests a role for an infectious agent yet to be identified.

We thank Drs Devred, Prades, Lefebvre, Silvie, Marti, Martin, Ostyn, Fontaine, Lemaire, and Lemahieu for their participation in the study. This work has been presented in part at the 16th Journées Francophones d'Hépatogastroentérologie (March 1992, Paris) and was supported by the Association F Aupetit, the Caisse Régionale d'Assurance Maladie de Nord-Picardie, the Ministère de la Santé et de l'Action Humanitaire (Direction Générale de la Santé), the INSERM, and Ferring Company.

TABLE II Children born to parents with inflammatory bowel disease

	Age (y)/sex	Age at onset	Type of IBD
B1	6/M 3/M		
C1	34/M 28/F 25/M 24/F	15 9 14 14	CD CD CD CD
C2	23/F	18	CD
C3	10/M 11/M 15/M 18/F		
C4	4/F 6/M		
C5	NA		
C6	16/M 18/F		
C7	NA		
C8	19/M		
C9	20/F 28/F		

NA = not applicable.

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