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What Factors Might Drive Voluntary Childlessness (VC) in Women with IBD? Does IBD-specific Pregnancy-related Knowledge Matter?

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Abstract

Introduction: Inflammatory Bowel Disease (IBD) affects many women of childbearing age, and rates of voluntary childlessness (VC) exceed those of the general population by far. The factors surrounding VC remain incompletely understood.

Methods: Female members of the patient organisation Crohn's and Colitis UK aged 18–45 years were invited to complete an online questionnaire collecting data on demographics, disease characteristics, Crohn's and Colitis pregnancy-specific disease-related knowledge (CCPKnow), and childlessness status.

Results: A total of 1324 women (mean age 33 years) completed the survey: 776 (59%) were diagnosed with Crohn's disease (CD), 496 (38%) with ulcerative colitis (UC) and 4% with inflammatory bowel disease–unclassified (IBD-U); 40% had children (14% pre-diagnosis (I); 26% post-diagnosis (II)), 36% planned to have children at some stage (III), 7% reported fertility problems (IV), and 17% were classified as voluntarily childless (VC). VC was associated with poorer CCPKnow scores [5.98 vs. 7.47 in (III); $p < 0.001$], older age [35 years old vs. 28 years old in (II); $p < 0.001$], unemployment (9.7% VC; $p < 0.001$), being single (34.5% VC; $p < 0.001$) not seeking medical advice ($p < 0.001$), and diagnosis of CD (19.3% vs. 13.9% UC; $p = 0.015$). Women with VC had more hospital admissions [mean 2.85 vs. 2.17 (III); $p = 0.03$] and surgical interventions [mean 1.27 vs. 0.65 (III); $p < 0.001$]

Conclusion: The aetiology of VC in women with IBD is multifactorial. Women's choice regarding children appears related to disease burden. VC is also associated with poor knowledge (CCPKnow), and women may stay childless unnecessarily. Patient education programmes could help to reduce the rate of VC in women with IBD, through correcting misconceptions and alleviating patient concerns.

Key Words: Inflammatory bowel disease; pregnancy; fertility; voluntary childlessness

1. Introduction

Inflammatory bowel disease (IBD), comprising Crohn's disease (CD), ulcerative colitis (UC), and IBD-unclassified (IBD-U), can manifest at any age.[1] IBD, therefore, affects many women of childbearing age.[2] Complex medical issues can arise for women with IBD who are considering pregnancy, and decision-making should be based on balancing the risks of treatment versus untreated disease.[2] Women often avoid medication during conception and pregnancy.[3,4] However, medication avoidance may lead to uncontrolled disease, which, in turn, poses a risk to mother and foetus (e.g. inflammatory burden

associated with low birth weight, premature birth, and even miscarriage or stillbirth).[5–9] Guidance with regard to the required complex decision-making is best placed with gastroenterologists because they have significantly better pregnancy-specific knowledge of IBD than obstetricians and general practitioners.[10]

IBD can adversely affect female fertility. However, most women with UC experience normal fertility,[11–13] and women with CD have lower fertility during disease flares only.[12,14–18] On the other hand, a history of IBD-related surgery, especially if the deep pelvis was involved or an ileo–anal pouch was formed, reduces fertility.[13,16,19–25] Overall fertility is, however, only marginally reduced [relative risk 0.99; 95% confidence interval (CI): 0.95–1.03], as demonstrated in a recent population-based study of 9635 women from the UK.[26] In contrast to the relatively small increase in infertility (involuntary childlessness), many women with IBD decide against having children (voluntary childlessness, VC). The rate of VC approached 17% in a US study, which greatly exceeded the VC rate of the general population.[27] There are no reliable data on VC from other countries (UK national statistics do not record the reason for childlessness) or health care settings. Previous studies have suggested that poor IBD-specific pregnancy-related patient knowledge and patient attitudes being in conflict with medical guidance are the main reasons for VC.[3,4,27,28] Selinger et al. have recently developed and validated a self-administered IBD and pregnancy-related patient knowledge assessment tool.[29] It has been shown that women with IBD who have children have better IBD and pregnancy-related knowledge than those without children.[29,30] The only study to look at VC and IBD-specific pregnancy-related patient knowledge found that women actively pursuing pregnancy had better knowledge than those with VC, but that study did not investigate the knowledge of the majority of childless patients who were not actively planning pregnancy at the time of the study.[31] Since it is likely that IBD pre-pregnancy counselling for women pursuing pregnancy and/or care received during pregnancy influence IBD and pregnancy-related patient knowledge positively, it is important to test the hypothesis that VC is associated with poor knowledge by comparing women with VC to those without children who are considering having children at some stage in the future. The previous analysis of those actively pursuing pregnancy at the time of the study risks overestimating IBD-specific pregnancy-related patient knowledge because these patients were more likely to have sought medical advice.[30]

The aim of this study was to ascertain the rate of VC in a large cohort of British women with IBD and to test the hypothesis that VC is associated with poor patient knowledge of IBD-specific pregnancy-related issues.

2. Methods

The study was performed using an online survey system (Bristol Online Surveys, UK) and included female patient members of Crohn's and Colitis UK: the largest UK charity providing peer support, patient education, and research funding with regard to IBD. We approached all 4300 female members aged 18–45 years who had previously supplied their email address to Crohn's and Colitis UK with an invitation to complete our online questionnaire. Each received two reminder invitations.

Patients' self-reported personal data included demographics, educational achievements, employment status, marital status and sexual orientation. Data on disease characteristics consisted of diagnosis, duration of illness, hospitalizations, surgical resections, and medication history. Medication adherence was assessed using the four-question version of the validated Medicine Adherence Report Scale (MARS).[32,33] Childlessness status and patient views were assessed using the methodology from the previous study by Marri, as follows.[27]

Women were placed in one of five categories according to their childlessness status:

I. Women with children born before diagnosis of IBD
II. Women with children born post-diagnosis
III. Women planning to have children at some stage
IV. Women experiencing fertility problems for at least a year
V. Women choosing VC

VC was defined as meeting all three of the following criteria:

a) Patient states that she had decided against having children
b) Patient had no children prior to or after diagnosis of IBD
c) Patient reported no fertility issues.

Disease-related pregnancy knowledge was recorded with the validated Crohn's and Colitis Pregnancy Knowledge Score (CCPKnow).[29] This 17-item questionnaire rates knowledge as poor (0–7), adequate (8–10), good (11–13), and very good (14–17). Participants were asked their reasons for choosing VC through the questionnaire by Marri et al.[27]

For the sample size calculation we estimated that, in accordance with previous studies, 15% of responders will be voluntarily childless and 20% will be considering having children in future. A sample size of 800 respondents (120 with VC and 160 considering having children) would give 90% power at 5% significance level to detect a 1-point difference in CCPKnow scores between women choosing VC versus those planning to have children.

Data analysis was performed with SPSS 22 (IBM, Armonk, USA) using ANOVA and the Student *t*-test for continuous data and Chi-square tests for categorical data with a *p*-value <5% interpreted as significant. The study was approved by the National Research Ethics Service Committee North West – Preston (14/NW/1391). Submission of the completed questionnaire was taken as informed consent.

3. Results

The survey was completed by 1324 women, with a mean age of 33 years (range 18–45 years) and a response rate of 31%. Of these, 76% were in a long-term relationship, 20% single, and 4% separated. Sixty-six participants (5%) identified themselves as being in a same-sex relationship. The majority of participants were either in employment (78%) or full-time education (8%). Further patient demographics are detailed in Table 1. Of the 1324 participants, 776 (59%) suffered from Crohn's disease (CD), 496 (38%) from ulcerative colitis (UC) and 52 (4%) from IBD-U. Hospital admission was previously required in 923 (69.3%) cases, and 437 (32.3%) of the cohort required previous resection surgery. Further disease and treatment characteristics are detailed in Table 2.

Of the 1324 participants, 524 (40%) had children [189 (14%) pre diagnosis (I); 335 (26%) post diagnosis (II)]. A further 480 women (36%) planned to have children at some stage (III). While in total 267 participants reported fertility problems, only 94 (7%) met the actual criteria for infertility (IV). An additional 226 women (17%) were classified as voluntarily childless (VC, Table 3). Patients reporting fertility problems, but not meeting the criteria for infertility, were categorized into groups I ($n = 46$), II ($n = 96$), III ($n = 24$), and VC ($n = 7$; patients had tried for pregnancy <1 year before deciding for VC).

Miscarriages had occurred for 283 (10.4%) women, while 142 women (10.4%) had experienced premature delivery. Fourteen women (1%) had adopted children. Just over half of the participants

Table 1. Patient demographics.

Age	Mean 33.5 years	Range 18–45 years
Ethnicity	White	1258 (95%)
	Asian	34 (2.6%)
	Black	6 (0.5%)
	Other	7 (0.5%)
Highest educational achievement	Secondary school	157 (11.9%)
	Apprenticeship/Guild/NVQ	386 (29.2%)
	Bachelor	577 (43.6%)
	Master/PhD	204 (15.4%)
Employment status	Full-time employment	697 (52.6%)
	Part-time employment	337 (25.5%)
	Full-time education	102 (7.7%)
	Unemployed	80 (6%)
	House person	108 (8.2%)
Relationship status	Single	259 (19.6%)
	Separated/divorced	44 (3.3%)
	Long-term relationship/married/civil partnership	1009 (76.2%)
Same-sex relationship	Yes	66 (5%)
	No	1250 (94.4%)
	Chose not to answer	8 (0.6%)

NVQ, National Vocational Qualifications.

Table 2. Disease and treatment characteristics.

Diagnosis	CD	776 (58.6%)
	UC	496 (37.5%)
	IBD-U	48 (3.6%)
	unknown	4 (0.3%)
Age at diagnosis		mean 25 years old
		median 24 years old
Duration of disease		mean 8.5 years
		median 7 years
Hospital admissions	Yes	923 (69.3%)
	No	401 (30.3%)
		Median 2 admissions
Bowel resection surgery	Yes	437 (32.3%)
	No	897 (67.7%)
		Median 2 resections
5-ASA medication	Current	615 (46.4%)
	Previous	521 (39.4%)
	Never	188 (14.2%)
Corticosteroids	Current	175 (13.2%)
	Previous	948 (71.6%)
	Never	201 (15.2%)
Immunomodulators	Current	597 (45.1%)
	Previous	368 (27.7%)
	Never	359 (27.1%)
Anti-TNF agents	Current	280 (21.1%)
	Previous	196 (14.8%)
	Never	848 (64.1%)
Adherence (MARS)	Mean	17.5
Scores >16 = adherent	Median	18
Score ≤ 16 = non-adherent	Score ≤16	280 of 1057 on therapy (26.4%)

CD, Crohn's disease; UC, ulcerative colitis; IBD-U, inflammatory bowel disease – unclassified; 5-ASA, 5-aminosalicylic acid; Anti-TNF, anti-tumour necrosis factor; MARS, Medicine Adherence Report Scale.

(673; 50.8%) had previously discussed pregnancy and IBD with one or more health care professionals [General Practitioner 300 (25.7%), Gastroenterologist 479 (41.1%), IBD nurse 263 (22.6%), Other 124 (10.6%)].

VC was associated with poorer pregnancy-specific disease-related knowledge (ANOVA, $p < 0.001$, Table 3). Knowledge levels were low for women choosing VC and those having had children prior to diagnosis. Most importantly, women choosing VC had much lower pregnancy-specific disease-related knowledge than those planning to have children in the future (CCPKnow 5.98 vs. 7.47 in (III); $p < 0.001$). VC was also associated with not seeking medical advice (25% vs. 9.4% for those seeking advice; Table 4; $p < 0.001$), and seeking medical advice was associated with lower VC, regardless of the consulted health care profession.

Participants choosing VC were older than women who had children post diagnosis (35 years of age vs. 28 years of age in (II); $p < 0.001$), were more likely to be single (34.5%; $p < 0.001$), and were more likely to be unemployed (9.7%; $p < 0.001$). Being in a same-sex relationship was not associated with VC. Older age at diagnosis (25.6 years of age) compared with patients planning for children at some stage (21.9 years of age (III); $p < 0.001$) was significantly associated with VC. There was, however, no difference in age at diagnosis compared with women with children post diagnosis (II, 24.1 years of age) or infertile patients (IV, 26.4 years of age).

Patients with a diagnosis of CD had higher levels of VC than those with a diagnosis of UC (19.3% vs. 13.9% UC; $p = 0.015$). Women choosing VC had more hospital admissions than women planning for children (mean 2.85 vs. 2.17 (III); $p = 0.03$), but not compared with women with children post diagnosis (mean 2.7; $p = 0.59$); had experienced more surgical interventions (mean 1.27 vs. 0.65 (III); $p < 0.001$) than women who were planning to have children at some stage and than those with children post diagnosis (mean 0.83; $p = 0.05$). When analysed as events per year of disease duration, however, the differences in surgical interventions and hospital admissions between women with VC and group III were no longer statistically significant, (Table 5). In contrast, exposure to different classes of IBD medication was not associated with VC (Table 5). Medication adherence was also not associated with VC.

Some participants with VC expressed concerns regarding pregnancy and IBD that focused on inheritance ($n = 152$), the ability to cope with raising a child ($n = 152$), and the influence of pregnancy on IBD ($n = 133$). Few patients reported to have been medically advised

Table 3. Child status and CCPKnow.

Group	Description	*n* =	CCPKnow
Group I	Children pre diagnosis	189 (14%)	5.59
Group II	Children post diagnosis	335 (26%)	8.89
Group III	Planning children at some stage	480 (36%)	7.47
Group IV	Infertile	94 (7)	7.55
VC	Voluntarily childless	226 (17%)	5.98

n, number; CCPKnow, Crohn's and Colitis pregnancy-specific disease-related knowledge. ANOVA $p < 0.001$.

Table 4. Association between seeking medical advice, CCPKnow and childlessness status.

Advice source	Used	Not used	
General practitioners re CCPKnow	n = 300 mean 8.44	*n* = 1024 mean 6.98	$p < 0.001$
General practitioners re childlessness status	I *n* = 24 II *n* = 123 III *n* = 69 IV *n* = 61 VC *n* = 23	I *n* = 165 II *n* = 212 III *n* = 411 IV *n* = 33 VC *n* = 203	Chi-square $p < 0.001$
Gastroenterologist re CCPKnow	*n* = 479 mean 9.18	*n* = 845 mean 6.24	$p < 0.001$
Gastroenterologist re childlessness status	I *n* = 33 II *n* = 206 III *n* = 154 IV *n* = 48 VC *n* = 38	I *n* = 156 II *n* = 129 III *n* = 326 IV *n* = 46 VC *n* = 188	Chi-square $p < 0.001$
IBD Nurse re CCPKnow	n=263 mean 9.27	*n* = 1061 mean 6.83	$p < 0.001$
IBD Nurse re childlessness status	I *n* = 19 II *n* = 88 III *n* = 15 IV *n* = 39 VC *n* = 102	I *n* = 207 II *n* = 392 III *n* = 174 IV *n* = 55 VC *n* = 233	Chi-square $p < 0.001$

Group I, children pre diagnosis; Group II children post diagnosis; Group III plan to have children at some stage; Group IV reported fertility problems; VC, voluntarily childless.

against pregnancy (*n* = 12), and few expressed concerns regarding potential birth defects (*n* = 41) or the potential for miscarriage/still birth (*n* = 29). Thirty-seven women reported remaining childless due to being sexually inactive.

4. Discussion

In addition to the classical treatment aims for IBD (to establish symptom control and avoid future complications), physicians also target restoration of quality of life for their patients.[34–36] For women of child-bearing age, quality of life should include the ability to have children, should they wish to do so. Marri's previous study has, however, reported that the rate of VC is much higher in women with IBD compared with the general population.[27] We acknowledge that VC can be a positive decision. Our concern is with regard to the potential for VC to be an *uninformed* decision, and although the reasons for VC in women with IBD have not been firmly established, higher burden of disease and poor pregnancy-specific disease-related patient knowledge leading to adverse attitudes have been proposed as potential explanations.[4,28] This study is the largest exploration to date of VC in women with IBD and the first to determine potential association between VC and pregnancy-specific disease-related patient knowledge in a British sample.

Our sample allowed us to separate participants by childlessness status into five different categories without losing statistical power. The ability to compare VC in different groups of women provides for clinically more meaningful comparisons than assessment of dichotomous outcomes (VC/not VC). For example, pregnancy-specific disease-related knowledge is best assessed against women planning to have children as opposed to those who have already had children post diagnosis, because their knowledge will likely have increased in the process of their care during pregnancy.

This study confirms that VC is a common occurrence in IBD, and our results slightly exceeded the level of VC previously reported.[27] Unfortunately, there were no data available on the extent of VC in the general population because the UK Office for National Statistics does not record the reasons for childlessness in their reports.[37] We have also established a number of factors associated with VC that help explain the phenomenon.

Our results are consistent with the hypothesis that VC is related to poor patient knowledge of IBD and pregnancy. The absolute difference in CCPKnow scores compared with women planning children at some stage (5.98 vs. 7.47) is of a magnitude (CCPKnow of group III are 25% higher than those of women with VC) that suggests serious information deficits, and previous work suggests that poor knowledge and negative patient attitudes towards pregnancy in contrast to medical evidence are closely related.[4] It is not surprising that women who already had children prior to diagnosis had lower CCPKnow scores in comparison with women who had children post diagnosis. These differences in pregnancy-specific disease-related knowledge likely reflect the fact that women who have completed their child planning prior

Table 5. Factors associated with voluntary childlessness.

Factor	Groups		Significance
Knowledge		CCPKnow	ANOVA $p < 0.001$
	I	5.59	VC vs. III $p < 0.001$
	II	8.89	
	III	7.47	
	IV	8.89	
	VC	5.98	
Consulting healthcare professionals		Spoken to professional	ANOVA $p < 0.001$
	I	55 (29.1%)	VC vs. III $p < 0.001$
	II	253 (75.5%)	
	III	217 (45.2%)	
	IV	85 (90.4%)	
	VC	63 (27.8%)	
Age		Years	ANOVA $p < 0.001$
	I	38.81	VC vs. III $p < 0.001$
	II	37.17	
	III	27.69	
	IV	36.29	
	VC	34.96	
Age at diagnosis		Years	ANOVA $p < 0.001$
	I	33.02	VC vs. III $p < 0.001$
	II	24.10	
	III	21.94	
	IV	26.44	
	VC	25.60	
Relationship status		Not in relationship	ANOVA $p < 0.001$
	I	11 (5.8%)	
	II	12 (3.5%)	
	III	153 (31.9%)	
	IV	5 (5.3%)	
	VC	78 (34.5%)	
Employment status		Unemployed	ANOVA $p < 0.001$
	I	15 (7.9%)	
	II	6 (1.8%)	
	III	29 (6%)	
	IV	8 (8.5%)	
	VC	22 (9.7%)	
Same-sex relationship		n =	ANOVA $p = 0.09$
	I	10 (5.3%)	
	II	10 (2.9%)	
	III	30 (6.3%)	
	IV	6 (6.4%)	
	VC	10 (4.4%)	
Diagnosis		VC: n =	ANOVA $p = 0.04$
	CD	150 (19.3%)	CD vs. UC $p = 0.015$
	UC	69 (13.9%)	
	IBD-U	5 (10.4%)	
	unknown	2 (50%)	
Hospital admissions		Mean	ANOVA $p < 0.001$
	I	1.89	VC vs. II $p = 0.59$
	II	2.70	VC vs. III $p = 0.03$
	III	2.17	
	IV	2.85	
	VC	2.85	
Hospital admissions per year of disease duration		Mean	ANOVA $p < 0.001$
	I	0.51	VC vs. II $p < 0.001$
	II	0.27	VC vs. III $p = 0.59$
	III	0.56	
	IV	0.33	
	VC	0.48	
Resection surgery		Mean	ANOVA $p < 0.001$
	I	0.51	VC vs. II $p = 0.05$
	II	0.83	VC vs. III $p < 0.001$
	III	0.65	
	IV	1.14	
	VC	1.27	

Table 5. *Continued*

Factor	Groups		Significance
Resection surgery per year of disease duration		Mean	ANOVA p = 0.001
	I	0.08	VC vs. II p = <0.001
	II	0.06	VC vs. III p = 0.13
	III	0.12	
	IV	0.09	
	VC	0.18	
Current 5-ASA prescription		*n* =	Chi-square p = 1
	I	105 (55.5%)	
	II	148 (44.1%)	
	III	221 (46.0%)	
	IV	42 (44.6%)	
	VC	99 (43.8%)	
Current immunomodulatory prescription		*n* =	Chi-square p = 0.03
	I	88 (46.6%)	VC vs. II p = 0.3
	II	142 (42.4%)	VS vs. III p = 0.5
	III	236 (49.2%)	
	IV	26 (27.7%)	
	VC	105 (46.5%)	
Current anti-TNF prescription		*n* =	Chi-square p = 0.18
	I	35 (18.5%)	
	II	58 (17.3%)	
	III	112 (23.3%)	
	IV	21 (22.3%)	
	VC	54 (23.8%)	
Current corticosteroid prescription		*n* =	Chi-square p = 0.14
	I	31 (16.4%)	
	II	38 (11.3%)	
	III	73 (15.2%)	
	IV	11 (11.7%)	
	VC	22 (9.7%)	
Medication adherence		MARS	ANOVA p = 0.091
	I	17.79	
	II	17.17	
	III	17.31	
	IV	17.57	
	VC	17.83	

CCPKnow, Crohn's and Colitis pregnancy-specific disease-related knowledge; Group I, children pre diagnosis; Group II children post diagnosis; Group III plan to have children at some stage; Group IV reported fertility problems; VC, voluntarily childless; CD, Crohn's disease; UC, ulcerative colitis; IBD-U, inflammatory bowel disease–unclassified; 5-ASA, 5-aminosalicylic acid; Anti-TNF, anti-tumour necrosis factor; MARS, Medicine Adherence Report Scale.

to diagnosis have little reason to learn about pregnancy and IBD, whereas the latter group may have received specific information during their pregnancy care. The mean level of CCPKnow for group III still falls under the category 'poor knowledge', suggesting that even in those thinking about pregnancy, further steps to improve pregnancy-related disease-specific patient knowledge should be considered.

The importance of pregnancy-specific disease-related knowledge is also underlined by the fact that seeking medical advice about IBD and pregnancy was associated with much lower levels of VC. Seeking medical advice may be related to the intention to have a child, but we have shown that this effect was independent of the profession of the health care provider consulted. It has previously been shown that gastroenterologists have a better knowledge of IBD and pregnancy than general practitioners and obstetricians,[10] but it appears that the information required to avoid VC can be provided by general practitioners. Very few patients with IBD have a contraindication to pregnancy. Reassuringly, very few patients reported to have received medical advice against pregnancy.

Our data also suggest that some women have chosen VC due to the burden of IBD. Women with CD had higher levels of VC than those with UC, and it is likely that a significant negative impact of IBD occurs more often in CD than in UC, given that some studies suggest that patients with CD experience lower quality of life than those with UC.[38] Our hypothesis, that a greater burden of disease is associated with VC, is further supported by data on hospital admissions and resection surgery. The VC group was associated with more hospital admission compared with women planning children, but not compared with those who had children post diagnosis, suggesting a modest effect of admissions on the decision to stay childless. Data on resection surgery are less ambiguous and clearly support the hypothesis. Moreover, it seems that surgery may have a greater effect on child-planning decisions than the experience of medical treatments. As women with VC had longer disease duration than group III, the effects of events per year of disease duration were also analysed and no significance was found. Events per year of disease duration could be seen as a more objective marker of disease severity, but it stands to reason that patients' perception of disease severity drives decisions regarding child planning. The cumulative disease experience, including overall number of hospital admissions and surgical resections, could influence the patients' perceptions more than the number of events per year of disease duration. In contrast, current exposure to different classes of IBD medication and adherence were not associated

with VC. Although medication decisions in the past (which were not assessed by this study) may have affected a patient's decision to have children, medications can often be switched, and reluctance to do so is potentially influenced by a desire to remain VC. We have previously demonstrated that general IBD-related patient knowledge does not influence medication adherence.[33] A higher burden of disease may influence women's decision-making in a number of ways: patients with a higher burden of disease may feel unable to cope with the added responsibility of looking after a child; some women may fear negative effects of pregnancy on their disease course; it is also possible that some women with a higher burden of disease chose VC due to poor pregnancy-specific disease-related knowledge, rather than due to making a well-informed decision.

The strengths of this study are the use of a relatively large sample of women with IBD from across the UK. Moreover, the disease distribution and severity in our sample reflects what is expected in UK IBD cohort studies. More women in our study were married or in long-term relationships compared with the UK adult general population.[39] Unemployment rates were similar to the general population[40], but more women in our group had degree-level qualifications.[41] Our cohort represents, in the authors' opinion, a group of women for whom decisions around having children are particularly relevant, given their response to the invitation to participate. Furthermore, the sample represents women more educated than the general population, which, if at all, would influence knowledge positively.

There are a number of limitations to our study. We relied on studying the members of a patient organization in order to recruit a large sample. However, patient organization membership has been shown to increase general disease-related knowledge[42] and CCPKnow scores [29] and thus led to a bias against supporting the knowledge hypothesis. The disease-related data were self-reported and could not be independently verified. It was, therefore, not feasible to collect in-depth details on surgical interventions. Ideally, we would wish to study prospectively the development of IBD- and pregnancy-related knowledge and decision-making processes regarding children over time in a defined cohort of women with IBD. Such a study would, however, be very costly and not provide results for a decade. It is, unfortunately, not feasible to follow our cohort longitudinally because the ethical approval required anonymity for all study participants.

Furthermore, it would be interesting to ascertain how individual women reach a decision for VC, but such intrusive and detailed questioning is outside the scope of an online questionnaire study. There is a relatively high level of educational achievement in our sample, and there is some evidence that women with higher education choose to have children later in life.[43] Within the constraint of the anonymous questionnaire (as required by the research ethics committee approval), we were unable to assess the effects of socioeconomic background, and health care utilization, or to conduct a follow-up study using the same cohort. We assessed intention to have children, and it is conceivable that some women may change their mind towards having children later in life. Relationship status often influences women's choice to have children. Selection bias in this domain is unlikely because the percentage of single women in our sample is close to the average in the general population.[39] We did not assign participants to groups according to current sexual activity because we felt that their childlessness status decision-making may not necessarily reflect current sexual activity. Previous sexual activity since diagnosis was not assessed in this study.

We considered a multivariate analysis using binary regression analysis to determine independence of associations, but decided against it. We compared VC versus groups II or III where more appropriate in univariate analysis. There are differences between groups II and III that would skew the results if were we to analyse the groups together or just choose one of them on binary regression analysis.

Comparison of VC rates with the general population or patients with functional bowel disease were unfortunately not possible due to the lack of relevant databases/registries.

5. Conclusion

The level of VC demonstrated in our cohort exceeds, in the authors' opinion, levels that can be explained by positive patient choice. This study suggests that women with IBD who chose to remain childless may benefit from increased knowledge about how their illness might be managed during and following pregnancy and to be supported by health care practitioners in making an informed decision in this regard. Even simple education strategies can improve knowledge significantly.[44] These could be administered by health care providers and patient organizations using multiple channels (e.g. information meetings, mail-outs, email, social media) to reach a wide audience of young women with IBD. Voluntary childlessness in IBD is associated with a relatively high burden of disease, and it is not surprising that that some women may approach motherhood with caution. However, many women with IBD may remain childless unnecessarily.

Conflict of Interest

The authors report no conflict of interest or funding.

Author Contributions

CPS designed the study, collected and analysed the data, and wrote the manuscript. JG contributed to data collection and analysis and critically reviewed the manuscript. AM contributed to data collection and analysis and critically reviewed the manuscript.

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