

Working Series 'Endometriosis in Wales: A comparative study on symptom awareness and help-seeking from the patient perspective for women living in Wales and other regions of the UK

J. Boivin, H. Jupp, H. Kingwell
School of Psychology, Cardiff University

Report published here: <http://psych.cf.ac.uk/contactsandpeople/boivin.php#two>

Introduction

Endometriosis is a common reproductive disorder affecting about 10% of women. Patients report that the provision of care for endometriosis is suboptimal, leading to very long delays in effective treatment. Delay in treatment could be due to numerous sources: patients could delay seeking help for worrying symptoms, doctors could delay the process of diagnosis, time to diagnosis or treatment could be lengthened due to healthcare system constraints, or to patient delay in uptake of treatment. The United Kingdom has a devolved healthcare system that means women seeking medical attention in any of its constituent countries (Wales, England, Scotland, Northern Ireland) could experience different barriers to effective treatment.

According to the Health Belief Model (Rosenstock, Strecher et al. 1988) diverse factors predict help-seeking behaviour. Faster presentation is associated with perceiving symptoms as threatening (e.g., symptoms perceived to be frequent, severe, worrying), reasons for seeking help outweigh reasons against seeking it, clear direct cues to get help exist (e.g., relative with disease encourages help-seeking) and person characteristics do not modify willingness to seek help (e.g., low disease knowledge, being older or non-white). Patterns of help-seeking are also influenced by the quality and organisation of healthcare. Care pathways that lack clarity, are difficult to comply with, or that do not meet patient need (or leave patients with unmet needs) can often compromise outcomes desired by patients and healthcare providers (e.g., efficient, effective).

The aim of the present mixed method study was to better understand experiences of symptoms and help-seeking in women with endometriosis living in Wales and other parts of the UK. To achieve this aim we undertook two studies. First, was a quantitative survey of women medically diagnosed with endometriosis recruited with the help of two patient advocacy groups (Fair Treatment for the Women of Wales, Endometriosis UK) and through university research panels. Women in Wales were compared to women in other parts of the UK and young women with pelvic or gynaecological symptoms not yet diagnosed. The second study collected qualitative experiential data in a one day arts-based workshop with 14 women living in diverse regions of Wales. This working paper reports the main findings of these studies.

Methods

Participants

The School of Psychology reviewed and approved the study protocol. The study protocol was developed in collaboration with two patient advocacy groups (i.e., Fair Treatment for the Women of Wales [FTWW], and Endometriosis UK). All participants were 16 years or older. Participants completed a quantitative survey or attended a workshop (or both). The number of women receiving the quantitative survey hyperlink and their eligibility for the study is unknown because women circulated onward through their networks and social media accounts. In total 2237 women clicked the survey link, 925 (41.4%, 925/2237) answered the questions relevant to the present report and of these 744 (80.4%, 744/925) pressed the 'submit' button at the end of answering the survey (a condition required by ethics committee to indicate consent). Women were invited to attend the Arts-based Workshop, and of these 14 were able to attend on the specified date.

Quantitative survey

a. Endometriosis group (N=569)

Participants with endometriosis (**Endometriosis Group**) were recruited via two patient advocacy websites (i.e., Fair Treatment for the Women of Wales [FTWW], and Endometriosis UK). Incentives for participation were not offered. Participants were assigned to the Endometriosis Group if they reported being diagnosed with endometriosis by a medical doctor. The Endometriosis Group was assigned to Wales (n=136) or Other UK (n=433), according to the reported place of residence.

b. Pelvic or gynaecological (PG) symptom group (N=175, comparison only)

A comparison group of female university students experiencing 'pelvic or gynaecological symptoms' were recruited from a Welsh university. Participants were invited through the School of Psychology Experiment Management System (EMS) and students that participated in exchange for credits. None of the women had been medically diagnosed with endometriosis. Participants were currently living in Wales due to their studies.

Qualitative workshop: Welsh women medically diagnosed with endometriosis (N=14)

Women from the advocacy group FTWW were invited to attend a one-day drawing workshop to provide more in-depth information about their symptoms and help-seeking.

Materials

This mixed methods study utilised two methodological approaches to collect data on the experience of symptoms and help-seeking, and these are described in the following sections. See Appendix 5.a. for more detailed information about methodology.

The 'Symptom and Help-seeking Survey' was created for the present study. The survey was similar for the Endometriosis and Pelvic & Gynaecological group but wording and questions were adapted to be relevant for each group. After agreeing to participate in the study participants confirmed whether or not a medical doctor had diagnosed their symptoms with endometriosis. Those who declared 'yes' were assigned to the Endometriosis group and completed the 'Symptom and Help-seeking Survey' in relation to endometriosis symptoms and help-seeking. Those who declared 'no' completed this questionnaire in relation to 'pelvic or gynaecological symptoms'.

‘Symptom and Help-seeking Survey’ was based on the Health Belief Model, which is a theoretical model to explain help-seeking behaviour. The questions were related to symptom burden (number, frequency, perceived severity, and worry about symptoms), (b) cues to support action (e.g., relatives with endometriosis, endometriosis knowledge), (c) importance of pros and cons of seeking medical advice, and (d) modifying individual factors that could increase or decrease likelihood of taking action (e.g., age, ethnicity, country of residence) and (e) help-seeking behaviour in particular the age at which they had started worrying about symptoms, sought medical advice, received a diagnosis, and were treated (see “Study variables”, Appendix 5a). The help-seeking section also comprised questions about experiences when first consulting the GP for symptoms (e.g., number of visits, referrals). Overall perceptions of care so far were also solicited. At the end of the survey participants were asked socio-demographic questions (e.g., age, education).

The arts-based workshop (‘Drawing Out Health’, Gameiro et al. website) is based on the premise and existing literature showing that drawing is a particularly effective method to elicit thoughts and feelings about sensitive topics (such as health) especially when personal experiences may otherwise impact the ability to express experiences of health (e.g., medical complexity, minority status, health literacy). Briefly, the one-day DrawingOut Health workshop (described in detail in Appendix 5.a) involves a set structure where women are taught basic drawing techniques, asked to draw around specific themes relevant to the health topic, present and discuss their drawings with the group (if willing), after which key messages from the group are compiled into a booklet for dissemination by women, and public engagement (anonymous). Advantages of this methodology include it being more inclusive than verbal-only methods, being unstructured, open-ended and non-directive allowing for highly personal perspectives to emerge, sharing experiences that enable central themes to be developed, and outputs (booklet) that facilitate broader sharing and engagement about the health topic outwith the group (Gameiro et al.). Three drawing activities were used to collect data. First, women were asked to **draw a self-portrait** and share it with the group. Second, participants were asked to **draw their symptom experiences**: ‘If endometriosis was an object, creature or animal or weather, what would it be?’). Third, participants were asked to **draw their experiences of healthcare**: ‘How would you draw your relationship with medical staff?’ After each exercise participants shared their artwork with the group, followed by group discussion of emerging views. Three researchers facilitated the workshop (rationale for research, DrawingOut Health methodology, drawing techniques) and the workshop audio recorded. Recordings were transcribed and analysed see Appendix 5.a for full details).

Data analysis

Descriptive statistics (percentages, means) and independent inferential tests (t-tests, analysis of variance) were used to compare groups on quantitative survey data. Thematic analysis was used for qualitative data elicited from the workshop according to well- established analytic approaches. Details in Appendix 5.a.

Results: Quantitative survey

1) Participant characteristics

Table 1 shows characteristics of the study groups. The Endometriosis groups (Wales, Other UK) were similar in age and educational background. The Wales Endometriosis group was more likely to be British, in a relationship and to have given birth (versus Other UK, PG groups). Although the Endometriosis groups were equally likely to be in full time work, more women in Wales were unemployed (20.0% versus 11.1%), and more reported poor or very poor health (42.6% versus 29.8%).

Table 1 Descriptive statistics for demographic characteristics in the Endometriosis Group according to country of residence¹

Variable	Wales (n=136)	Other UK (n=433)	Pelvic & Gynaecol (n=175)	Statistic
Age, years M (SD)	34.87 ^a (7.63)	34.35 ^a (8.33)	26.27 ^b (8.6)	64.74** *
Age groups n (%)				
19 - 25	16 (12.3)	62 (14.7)	101 (58.4)	
26 – 40	87 (66.9)	264 (62.4)	58 (33.5)	
41 to 70	28 (20.8)	96 (22.7)	14 (8.1)	
Ethnic origin n (% White British)	129 (94.9) ^a	382 (88.2) ^b	145 (82.9) ^b	10.6**
University Education n (% yes)	73 (53.7) ^a	264 (61.0) ^a	----	2.3
Employment n (%)				9.39*
Full time	73 (53.7) ^a	254 (58.7) ^a	----	1.05
Unemployed	27 (20.0) ^a	48 (11.1) ^b	----	6.95**
Relationship status n (%)				
In a relationship (married, cohabitating, together apart)	124 (91.2) ^a	349 (80.6) ^b	121(69.1) ^c	23.5***
Ever given birth n (% yes)	65 (47.8) ^a	164 (37.9) ^b	37 (21.1) ^c	4.2*
Self-reported health n (% poor or very poor health)	58 (42.6) ^a	129 (29.8) ^b	40 (22.9) ^c	14.4 ***

Note. ¹Ns vary slightly due to isolated missing data. Values (means, percentages) with different superscript letters are significantly different from each other. * p < .05

** p < .01

*** p < .001

2) Symptom profile of endometriosis versus other pelvic and gynaecological symptoms groups

Table 2 shows descriptive statistics for symptom profiles. The Endometriosis groups recalled their symptoms starting at a similar age (early 20s) and a similar percentage (about 15%) had a mother or sister with endometriosis. In terms of symptom profile, women in Wales reported a greater number of symptoms, a greater percentage experienced daily, and more severe symptoms than the Other UK group and the PG group. The two Endometriosis groups (Wales, Other UK) had a greater symptom burden in all respects (number, frequency, severity) than did the Pelvic & Gynaecological group, except for worry about the symptom that was less in the Endometriosis group. **Table 3** shows the percentage of women reporting any experience of each symptom in the symptom profile list (generated through research and advocacy groups). The comparison tests show that a major distinguishing feature was that more women in the Endometriosis group reported daily experience of symptoms than did women in the comparison group of undiagnosed pelvic and gynaecological symptoms.

Table 2 Descriptive statistics for symptom profile for the Endometriosis Group according to country of residence

Variable	Wales (n=136)	Other UK (n=433)	Pelvic & Gynaecological (PG) (n=175)	Statistic
<i>Age of onset and family background</i>				
Average age at symptom start	21.60 (5.66) ^a	22.87 (6.37) ^a	20.0 (6.1) ^b	12.60*
Mother, sister with endometriosis n (%)	21 (15.4) ^a	52 (12.0) ^a	40 (22.9) ^b	11.40**
Any other female relative with endometriosis n (%)	78 (57.4) ^a	214 (49.4) ^b	75 (42.9) ^b	6.4*
<i>Symptom profile</i>				
Nbr of symptoms M (SD)	10.18 (3.1) ^a	9.22 (3.1) ^b	7.41 (3.3) ^c	33.43**
Total Symptom burden M (SD)	173.55 (77.0) ^a	151.64 (75.66) ^b	122.9 (69.5) ^c	17.5***
% symptoms daily M (SD)	.51 (.25) ^a	.43 (.27) ^b	.29 (.26) ^c	29.89***
Mean symptom severity M (SD)	3.89 (.56) ^a	3.71 (.59) ^b	3.34 (.70) ^c	37.08***
Mean symptom worry M (SD)	2.20 (.67) ^a	2.33 (.76) ^a	2.88 (.94) ^b	37.09***
<i>Self-reported time intervals and doctor visits</i>				
Total time interval between worrying symptom awareness and treatment (in years) M (SD)	9.43 (7.1)	8.50 (6.87)	-	1.30
Symptom awareness to seeking advice	1.66 (3.21)	1.63 (3.92)	-	.07
Seeking advice to diagnosis	7.56 (7.18)	6.53 (6.48)	-	1.56
Diagnosis to treatment	.52 (1.76)	.32 (1.33)	-	1.38
Estimated number of doctor visits before diagnosis M (SD)	26.0 (34.1)	19.89 (32.0)	-	1.79
Nbr more than 20 visits n (%)	51 (37.5)	115 (26.6)	-	5.86*
<i>Reported thoughts and feelings about diagnostic process</i>				
Emotional reaction to diagnosis				
Negative emotions	.391 (.22)	.373 (.22)	---	.76
Positive emotions	.142 (.14)	.166 (.16)	---	1.60
<i>Overall view of healthcare</i>				
Satisfaction with medical experiences	2.41 (.87)	2.46 (.87)	-	.56
How often feel like giving up	3.36 (1.4)	3.01 (1.38)	-	2.56*

Notes. * p < .05 ** p < .01 *** p < .001

Table 3

Percentage of women reporting experience of symptom according to country of residence and versus women experiencing pelvic and gynaecological symptoms not diagnosed as endometriosis

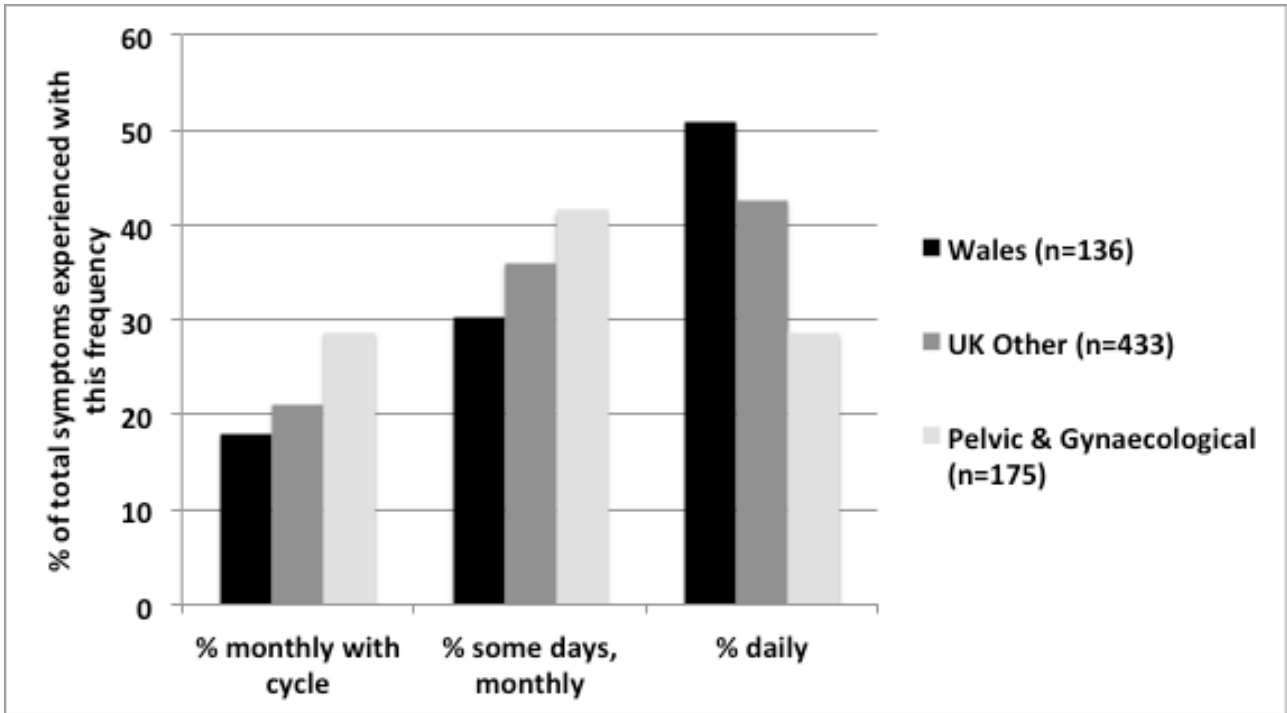
	Endometriosis		Pelvic & Gynaecological ¹		
	Wales (n=136)	Other UK (n=433)	Wales university students (N=175)	Chi value	Comparison tests ² E = Endometriosis PG = Pelvic & Gynaecological
Symptom	n (%)	n (%)	n (%)		
Period pain/affects daily activities & QoL	132 (97.1)	416 (96.1)	161 (92.0)	29.2***	E > daily: 41.2% vs 19.9%
Cyclical pelvic pain	134 (62.9)	308 (71.1)	29 (16.6)	10.5*	E > daily: 49% vs 34.9
Chronic pelvic pain (pain felt below your belly button) for at least 6 months	133 (97.80)	402 (92.8)	108 (61.7)	21.7***	E > daily: 72.2% vs 50.9%
Pain and discomfort when passing water	87 (64.0)	258 (59.6)	79 (45.1)	10.8*	E > daily: 38.9% vs 21.5%
Pain and discomfort during bowel movement	125 (91.9)	357 (82.4)	105 (60.0)	18.5***	E > daily: 60.6% vs 39.0%
Pain during or after sex	120 (88.2)	345 (79.7)	115 (65.7)	16.6**	E > daily: 60.6% vs 43.5%
Unexplained cyclical pain under ribcage	83 (61.0)	232 (53.6)	62 (35.4)	11.9*	Daily frequency reported: 39.2% Other UK; 51.8% Wales; 27.4% PG
Shoulder tip pain	69 (50.7)	167 (38.6)	49 (28.0)	4.4	
Difficulty emptying bladder	76 (55.9)	242 (55.9)	71 (40.6)	17.8***	E > daily: 48.5% vs 22.5%
Heavy periods (bleeding could soak through your clothes/using pads & tampons together)	119 (87.5)	345 (79.7)	137 (78.3)	5.6	
Blood in urine	46 (33.8)	133 (30.7)	31 (17.7)	10.1	Wales > daily: 28.3% vs 13.2%
Rectal bleeding	69 (50.7)	176 (40.6)	58 (33.1)	9.1	
Coughing up blood	9 (6.6)	22 (5.1)	2 (1.1)	1.1	
Breathlessness,	68 (50.0)	184 (42.5)	72 (41.1)	8.2	
Feeling tired all the time	129 (94.9)	397 (91.7)	150 (85.7)	12.31*	Wales > daily: 89.9% vs 77.0%

Note. ¹Pelvic or gynaecological (PG) = University students with pelvic or gynaecological symptoms not diagnosed as endometriosis. ²Frequency of experience 'monthly with period', 'some days, monthly', 'daily, most days'; 'E= Endometriosis group incl Wales and Other UK, QoL = Quality of Life

Figure 1 shows the difference in frequency of daily symptoms among groups. The Endometriosis groups (especially living in Wales) were more likely to experience daily symptoms, whereas the Pelvic and Gynaecological groups were more likely to have cyclical or less frequent symptoms.

Figure 1

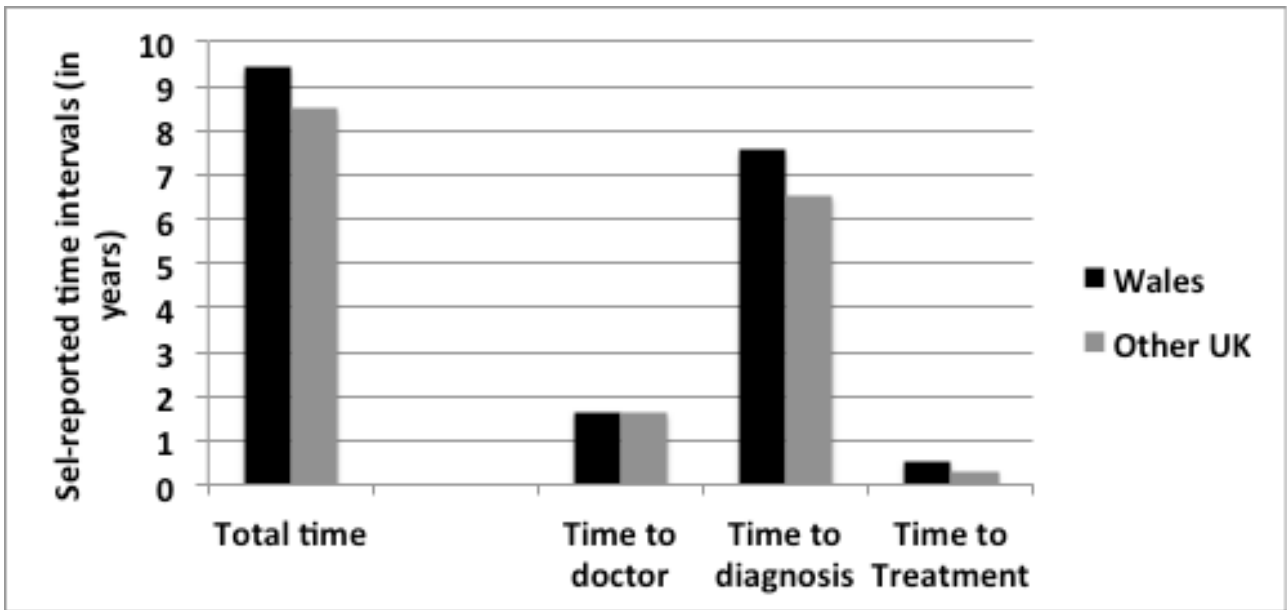
Percentage of total symptoms experienced at each frequency ‘monthly with period’, ‘some days, monthly’, ‘daily, most days’ according to group



3) Help-seeking and perceptions of provision of care

The Endometriosis groups were similar in helping seeking patterns with time intervals. **Table 2** shows that the total time interval between symptom awareness and treatment was 9.43 years for women in Wales and 8.50 years for women in the Other UK group. When this time delay was decomposed into its constituent elements, the longest reported time interval is between women presenting to the doctor and getting a diagnosis (see **Figure 2**, about 7.5 years in Wales versus 6.5 years in Other UK). Women in Wales reported more medical visits to achieve a diagnosis (26 versus 20 visits), with significantly more requiring 20 or more visits (20% reported more than 40 visits versus 12.2% in Other UK) (see **Table 2**). Note that 8.8% of women in Wales and 5.5% in Other UK reported having had too many visits to provide an estimate. Logistic regression analysis indicated that predictors of a longer diagnostic time interval were having an older age at start of symptoms, reporting negative experiences at first consultation (e.g., disbelief doctor, misdiagnosis). There was a trend for having a close relative with endometriosis (mother, sister) to be associated with a shorter time to diagnosis.

Figure 2: Total time interval between symptom awareness and first treatment, and decomposed into its constituent time intervals



Participants in the Endometriosis group reacted negatively to the diagnosis of endometriosis (see **Table 2**) but 29.2% felt validated by the diagnosis. The Endometriosis groups were similarly satisfied with their experiences of care thus far (seldom satisfied), but significantly more women in Wales than Other UK felt like giving up on having their symptoms explained or diagnosed. As shown in **Table 4**, the majority of participants in the Endometriosis groups believed that the doctor contributed to the delay in getting a diagnosis (about 80%). The Endometriosis groups were very similar in the reasons for believing this with the most frequently endorsed reasons being the doctor not believing the symptom profile (about 60%), being given medication that was not sufficient (60%) and misdiagnosis (through judgement or false negative testing).

Table 4 Number (percentage) of patients that believe doctor caused a delay getting a diagnosis and reasons for that belief according to Endometriosis group

Perceived reasons for doctor causing delay	Wales (n=136) n (%)	Other UK (n=433) n (%)	Chi square
Believe doctor caused a delay in getting a correct diagnosis n (%) yes	104 (78.2)	340 (80.4)	.30
<i>Reasons endorsed:</i>			
Doctor didn't believe the severity of my symptoms or thought it was normal pain	89 (65.4)	275 (63.5)	.68
The doctor thought my symptoms were due to mental health problems (for example, stress, anxiety or depression)	33 (24.3)	105 (24.2)	.00
The doctor said I was too young or too old to have endometriosis	28 (20.6)	51 (11.8)	6.71**
The doctor initially diagnosed me with something other than endometriosis	47 (34.6)	122 (28.2)	2.0
I had an ultrasound that failed to detect my endometriosis	46 (33.8)	143 (33.0)	.86
Abnormalities were detected but were not considered important or acted upon at the time of discovery	13 (9.6)	45 (10.4)	.08
I was first prescribed the oral contraceptive pill to relieve symptoms, but later needed alternative hormonal therapies	94 (69.1)	316 (73.0)	.38
I had to unnecessarily repeat tests that delayed the process (e.g., ultrasounds, pregnancy tests, sexually transmitted diseases)	34 (25.0)	92 (21.2)	.85
The doctor told me to get pregnant to reduce symptoms of endometriosis	25 (18.4)	70 (16.2)	.36
The doctor said they could not refer me because the system would not allow it)	13 (9.6)	23 (5.3)	3.1 [†]

Results: Qualitative workshop

Women in the Workshop were on average 34 years of age (4 < 30 years), 8 had an undergraduate or postgraduate degree and average years since diagnosis was about 8 years. All women were living in Wales.

The one-day workshop generated 4447 verbatim sentences that could be coded. The content of these sentences generated 119 codes. The researchers (HK and JB) grouped these 199 codes into 20 thematically related categories. In the final step of the analyses these thematic codes were abstracted into the four broad themes of the workshop. **Table 5** shows how codes were clustered into five broad themes, of which four were considered central to the workshop findings. **Figure 3** illustrates the four broad themes and their links.

Table 5 Thematic categories subsumed under each broad themes (in bold) for women's experiences of endometriosis (N=4447 coded elements)

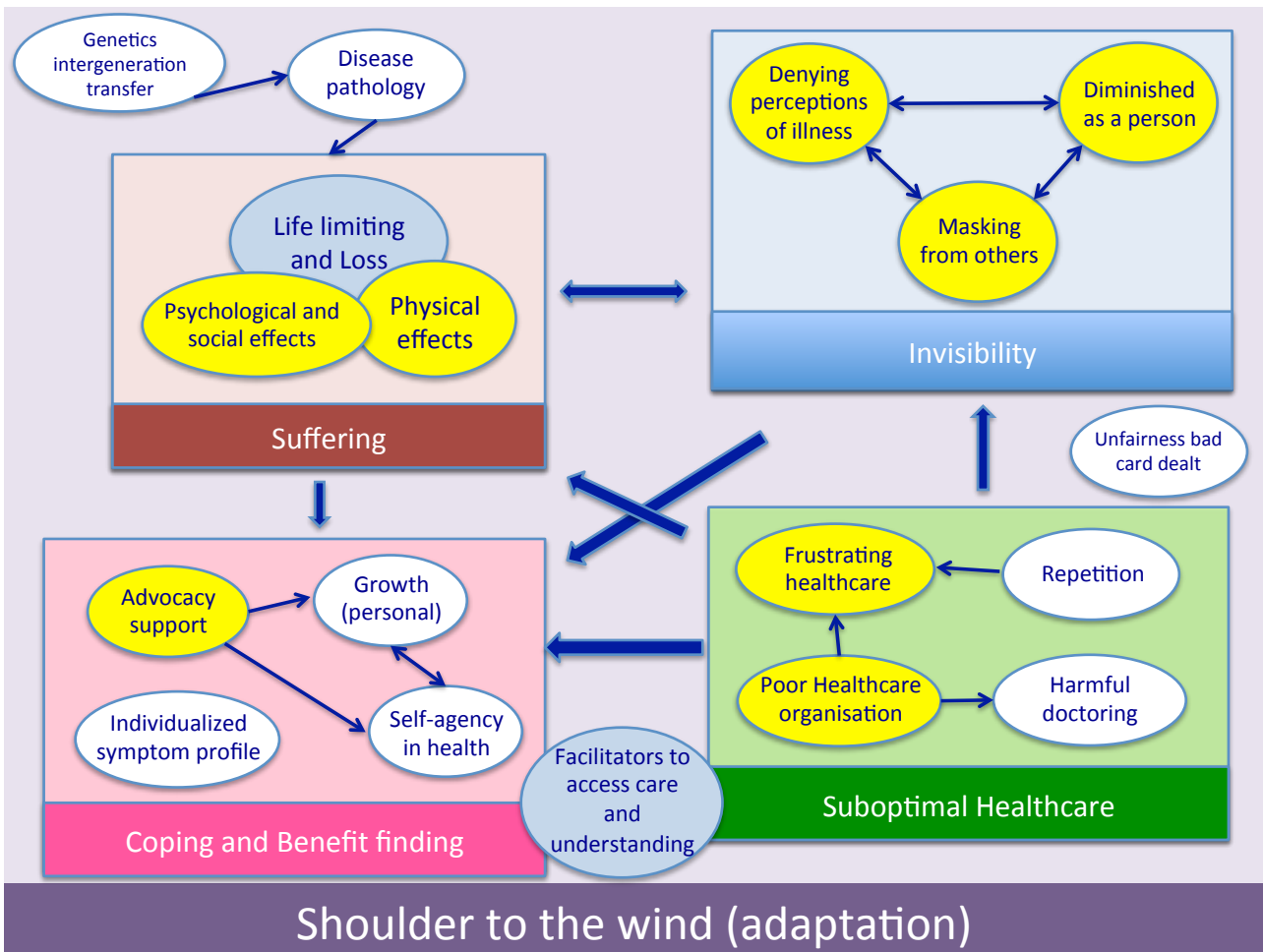
Broad themes	%	n
Underpinning thematic categories		
Disease reference	12.82	438
Disease pathology	11.89	417
Genetics intergeneration transfer	0.93	21
Suffering	41.92	1348
Psychological and social effects	19.29	675
Physical effects	11.41	324
Life limiting	5.21	172
Loss	6.01	177
Invisibility	23.38	776
Social perceptions of illness	16.87	607
Diminished as a person	4.92	124
Masking from others	1.59	45
Sup-optimal healthcare	35.36	1203
Frustrating healthcare	13.45	418
Poor healthcare organisation	18.35	693
Harmful doctoring	3.56	92
Coping and benefit finding	25.17	814
Advocacy support	11.47	408
Growth (personal)	5.53	156
Self-agency in health	3.29	107
Individualized symptom profile	4.88	143
Other	23.46	695
Facilitators to access care and understanding	2.31	47
Unfairness (resentment) bad card dealt	3.91	97
Poor health due to other factors	1.41	46
Issues emerging from research	3.01	67

n= number of times thematic category was mentioned by any participant

%= percentage of coded data (verbatim sentences) linked to thematic category

NB: The sum '%' and 'n' for the broad themes do not add up to 100% (i.e., n=4447) as some codes feature in more than one thematic category



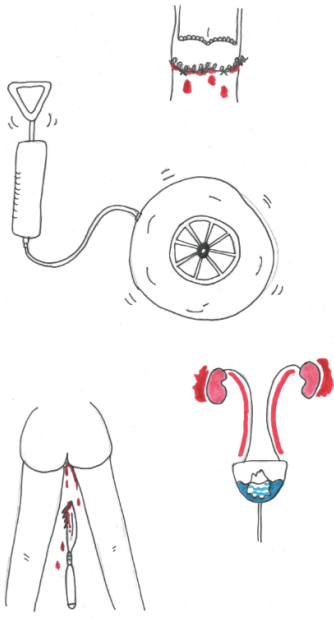
Figure 3 Thematic representation of the four broad themes to emerge from the arts-based DrawingOut Health workshop



The broad themes are briefly described in the next sections, with a full report forthcoming.


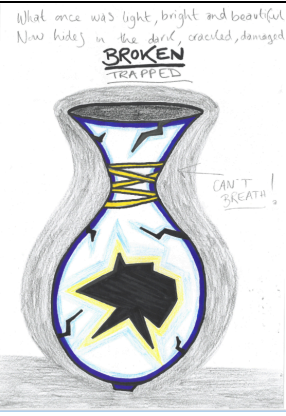
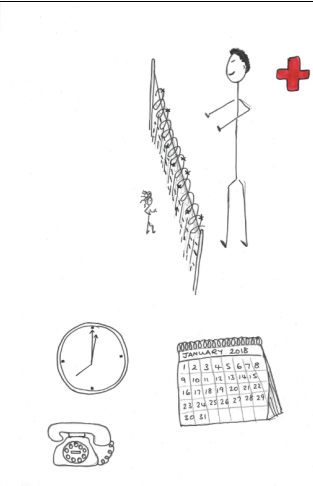
I. Endometriosis is suffering

As shown in Figure 3 and Table 5 much of the coded data referred in some way or another to the suffering women experienced as a result of their condition. The suffering was physical and psychological, considered to be life limiting and associated with significant loss. These quotes and drawings illustrate this suffering.

<p>IL: me the thing that like I just remember or just features really really heavily is heaviness (.) so I always feel dragged towards the ground in some way (.) so it was like- when I was looking at the bubbles and stuff I don't know it felt right that everything just sort of gets pulled down or just almost like melts down so everything has kind of- I tried to put that in the lettering where it's heavy at the bottom and everything just feels like oh just tugged downwards so that's what (.) that's the overarching like</p> <p>ED: that's almost like- that picture there is almost like you're being devoured by a monster</p> <p>LI: yeah and it's kind of (is)</p>	
<p>AD: my drawings are- there's two of them but they're both of me like curled in the foetal position crying (.) er and my thoughts are basically I can't take this anymore and I don't want to carry on like this (.) cos that's how it makes you feel (.)</p> <p>EJ: you're not alone in that</p> <p>RJ: I was gonna say how many- how many of us in here have considered suicide [show of hands] look at all the hands (.)</p>	
<p>um ...that's the barbed wire on the bra (.) cos I get a lot of pain upper abdominal pain and like under the ribs (.) that's a pump going to a tyre that's overinflated and it's gonna burst cos when you get bloating you literally feel like you're gonna pop (.) I don't know if you can tell what that is but it's an iceberg in my bladder cos I get this weird cold sensation symptom (.) ... yeah um that's a knife (.) sorry that's a bit graphic that's a knife going into the rectum...</p>	

II. Invisibility through the eyes of others and self

Three themes were linked to this broad theme. Women often expressed that because endometriosis was not visible others often would not believe in the severity of the illness. These denying social perceptions made it more difficult for people to talk about their disorders. Women often felt they had to present a brave face, deny the extent of their suffering or in other ways hide to meet with what they perceived were the (at times judgemental) perceptions of doctors, and other people in their social environment. Invisibility was also shown in the diminishing effect of patronising healthcare or of feeling diminished as women (less feminine, less able, less confident). This invisibility is illustrated in the following drawings and quotes.

<p>...you look perfectly normal nobody can see your scars; they're hidden away. Nobody can see your pain and you wear a mask all of the time you tell people that you're fine and everybody thinks that you're- (.) but really [sigh] that's how I really see myself that's how I look without my make-up that's how I feel about myself.</p>	
<p>EJ: ... a vase yeah ... what once was light bright and beautiful now hides in dark, cracked, damaged and broken because that is how I feel quite frankly. ... I put on my face and I put on my yellow bits and let you all see me smiling but if you saw me at home I don't look like this (.) I don't stand up straight I walk around my house like a granny with a hot water bottle shoved down the front of my pants down the back of my pants (.) I'm constantly trying to do anything I can to make myself feel better so that I can get back out and help other people feel better because (.)</p>	
<p>MS: ... um so I drew- that's the doctor (.) the big one there (.) the big stick and then that's me tiny little insignificant me and um a six foot high wall with razor wire and everything across the top of it</p>	



IV. Coping through benefit finding and shared experiences

Women managed to cope with their illness by taking their health and healthcare into their own hands. All expressed that an important way in which they were able to achieve this and cope was through the advocacy group. All the women in this workshop had been recruited through Fair Treatment for the Women of Wales but women expressed gratitude for other groups through the experiences they had had over the long years of coping with the illness.

AL: it's the best thing I've ever done and I really mean that joining FTWW.

RJ: I remember telling my husband I'm like oh my god this is the most amazing group ever

EJ: for me the only way I can become (not) so broken anymore is by trying to help fix the whole scenario that we're living in cos it's just not good enough

<p>YL: I wouldn't have started the Swansea group (support group) if I hadn't had the confidence that I got through finding FTWW I know that for certain</p>	
<p>ED: maybe at last our voices are going to be heard (.) I- um so the fact... that Welsh government listen to patients you know how amazing is that ... my god you know if this really does happen (change in provision for endometriosis in Wales) ... just imagine how wonderful it would be if Wales led the way on endometriosis</p> <p>AL... just the fact that this today is happening just the fact that new medics like Hannah (medical student facilitating the workshop) are going into the profession and are interested in and want to know is- is encouraging.</p>	

V. Over-arching theme: Shoulder to the wind (adaptation)

The overarching theme of the workshop referred to the adaptation the women made to carry on with life despite the significant suffering, invisibility afforded to their condition (and diminishing selves) and the inefficient, fragmented and at times chaotic healthcare to which they were exposed. This adaptation was achieved largely through self-agency and connecting together as women (through advocacy groups) and developing strong bonds of connection.

...when it's actually blowing a gale you're facing forward and trying to push forward through all of that with sort of like a bit of weight from there pulling you backwards and it's just like you've got to keep going cos where you need to be is over there but every force possible is pushing you back (.)



Conclusions

The results are compelling in showing that women with endometriosis living in Wales suffer greatly from their condition and this suffering is linked to the suboptimal healthcare they believe they receive. A limitation of the study is that participants with endometriosis were recruited through advocacy groups. It could be that women seeking out such groups have worse disease profiles or in other ways differ from those not linked to advocacy groups. We acknowledge this limitation and support further controlled research. However, we also note that > 500 women participated in the studies and that we used a comparison group of women with undiagnosed pelvic and gynaecological symptoms (175 women) which helped mitigate bias from sampling.

Participants in Wales were found to have a more severe disease burden (i.e., more symptoms, more frequently experienced and of greater severity), to require more consultations to achieve a diagnosis (26 visits) and to have a longer time to diagnosis (7 to 8 years) than women in other parts of the UK. It could be that greater disease burden means that women in Wales have more complex disease that is more difficult to diagnose. However, we would argue the reverse; greater disease burden is a consequence of the inefficient care pathway and long delay in diagnosis that allows the disease to progress to a worse state. The majority of participants in Wales and other parts of the UK reported unsatisfactory early medical consultations that minimised or normalised their symptoms, that subjected them to unnecessary repeat testing, and that often led to incorrect diagnoses or ineffective first treatments requiring further, more invasive treatments at a later date. Together these findings suggest that the current care pathway is not efficient, and attracts additional costs to the national health service in Wales through physical disease progression and through additional health services to manage suffering (e.g., depression, pain management) and potentially through other lost opportunities. Indeed participants living in Wales were two times more likely to be unemployed despite being as educated as their counterparts in other areas of the UK. Our results strongly support further investigation of the care pathways for endometriosis in Wales, more research into development of educational resources for doctors about endometriosis and young girls about menstrual health and development of support tools for women with endometriosis. The findings of the workshop made clear that women felt fortunate in finding support and information through advocacy groups that helped them strengthen their resilience and coping. Despite strong evidence of coping in the face of adversity care for women with endometriosis in Wales should be re-evaluated.

Appendix 5.a Detailed description of quantitative survey questions and qualitative drawing workshop

I. Quantitative survey

'Symptom and Help-seeking Survey' was based on the Health Belief Model, which is a theoretical model to explain help-seeking behaviour. It comprises factors predicting the help-seeking from the person perspective. These include: (a) symptom burden (susceptibility, severity to disease), (b) cues to support action, (c) pros and cons of taking action, and (d) modifying individual factors that could increase or decrease likelihood of taking action (e) help-seeking factors and (f) experiences of care.

a) Symptom burden

Symptom burden was the cross product of the frequency of symptoms, their perceived severity and extent of worry about the symptoms. The 15 symptoms were: period pain that affects daily activities and quality of life, heavy periods (bleeding so heavy it could soak through your clothes or using pads and tampons together), chronic pelvic pain (pain felt below your belly button) for at least 6 months, pain during or after sex, pain and discomfort when passing water, difficulty emptying bladder, blood in urine, pain and discomfort during bowel movement, rectal bleeding, unexplained cyclical pain under ribcage, cyclical pelvic pain. Four non-specific symptoms were also included on advice of advocacy groups: shoulder tip pain, breathlessness, coughing up blood, feeling tired all the time. Participants were asked to indicate frequency of each symptom, then grouped in three categories ('daily, most days', 'some days, monthly', 'monthly with my period' coded, 3, 2, 1, respectively). Physical severity ("How severely you experience each symptom?") and worry ("How worried are you about each symptom?") were each rated on 5-point response scales from 'not at all' to 'extremely' (1 to 5). Product scores (frequency X severity X worry) for each symptom were calculated and summed to produce a Total Symptom Burden score (range: 15 to 1125) for each person.

b) Cues to support action

The cues to action were factors that would increase perceptions that symptoms were worthy of seeking medical help. The factors were: having a mother or sister with endometriosis (no, yes), any other relative with endometriosis (no, yes), age at which symptoms started and actively trying to become pregnant (yes, no).

c) Pros and cons of seeking help

The reasons for or against seeking help were generated based on extant literature on reasons for help-seeking, discussion with advocacy groups and the recent NICE Guideline. There were 19 reasons for seeking help (e.g., desire to stop symptom, pain too severe) and 15 reasons against (e.g., too embarrassed, thought symptoms were normal). Participants rated to what extent these reasons had been important in motivating their decision about seeking medical help on a five-point response scale (strongly disagree to agree). Responses were averaged across items within each set (pros, cons) with higher scores indicating more of the attribute (more pro, more con). In addition, cons were subtracted from the pro score to create a facilitated help-seeking score (higher scores indicated pros outweighed cons).

d) Modifying factors

Four modifying demographic factors known to affect help-seeking were also collected and were: age, ethnic origin (British, non-British), country of residence (Wales, versus other-UK) and relationship status (in a relationship, not in a relationship) as well as knowledge about endometriosis. The Endometriosis Knowledge Scale (EKS) was created for this study and comprised eleven statements culled from existing research, key messages issued from advocacy groups, and content from the NICE guidelines (2017). The items included facts about endometriosis prevalence, symptoms and treatments. The Cronbach reliability

coefficient was satisfactory (.74 for the set of 11 symptoms). Questions were scored and a total correct score was converted to a percentage (0 to 100% correct). The Endometriosis group was additionally asked about what they thought could help detect endometriosis (laparoscopy, blood tests, scans, x-rays, pelvic exams, colonoscopy, MRI).

e) Help-seeking behaviour

Diverse aspects of seeking help were investigated. Delay was computed based on answers to four age questions, as per Bougale et al. (2017): age started thinking one or more of your symptoms were not normal; age went to see a doctor about your symptoms; age were given a diagnosis of endometriosis, and; age were first treated for endometriosis. From these variables we computed four time intervals: (1) **help-seeking interval** which was time between worrying symptom awareness and first consulting doctor, (2) **diagnostic interval** which was time between first consulting doctor and receipt of a diagnosis; (3) **treatment interval** which was time between diagnosis to receipt of first treatment [whether effective or not], and; (4) **total time interval** which was time between worrying symptom awareness and first treatment. Participants had the option to indicate that they had never been treated but all were diagnosed as a condition of the study. Time variables were expressed in [fraction] of years, with 0 indicating no time interval and > 0 indicating at least some [fraction] of years.

Although women in the **Pelvic Gynaecological Group** were asked these questions, too few had consulted a medical doctor about their symptoms (n=36, 20.6%) to analyse separately, and therefore they were excluded from analyses using the time interval variables. However, these women were asked **to what extent they felt susceptible to having endometriosis given their pelvic and gynaecological symptoms** (“How likely do you think it is that you have endometriosis compared to most other women your age?”, much more likely to much less likely, or don’t know). They were also asked about **how long it would take them to seek medical attention** (“If you had a symptom that you thought might be a sign of endometriosis, how long would it take you to go to the doctors from the time you first noticed the symptom?” adapted from Simon & Wardle, 2012). Participants indicated the number of weeks. Responses were re-coded as ‘0= I would go as soon as I noticed, no delay’ and ‘1= any delay, between up to a week to more than a month’ (based on Smits et al. e-cancer paper) [data for the latter two questions published in forthcoming report].

f) Experiences of care for endometriosis (Endometriosis group only)

Women were asked to inform on their experiences of care at the time of first consultation. First, were questions about the **number of medical consultations prior to diagnosis** (“estimate the number of visits you made to your doctor prior to your diagnosis of endometriosis”) and how easy or difficult it was to see a doctor (very difficult to very easy). Second, were questions about **what the doctor did at this first consultation** (“did the doctor explain what could be the cause of symptoms”, “did the doctor refer you for tests”, yes, no) and what were **tests in referral** (blood urine, sexually transmitted infections, x-rays, scans, including the option of not being referred for further testing). Third, were questions about the **emotional and cognitive reactions to diagnostic consultations**. Emotional reaction to diagnosis comprised 8 negative and 8 positive emotions, 4 uncertainty reactions, and 1 appraisal (validation) question suggested by advocacy group. Cognitions included **perceptions the doctor had caused a delay in diagnosis** (yes, no), and if yes **reasons for this belief** (e.g., normalised symptoms, first provided oral contraceptives but needed other hormonal preparations, had scan that failed to detect endometriosis, initial diagnosis was other than endometriosis, unnecessary repetition of test [e.g., scans, pregnancy tests, STI tests], recommended to get pregnant to reduce symptoms, symptoms were ascribed to mental health problems, not referred for tests due to constraints of system). Finally participants were asked to provide an over view of **satisfaction with medical care so far** by indicating how frequently they were satisfied with medical experiences thus far (“Taking all of these experiences together how often were you satisfied with your medical consultations”)

from (never, seldom, quite often, very often, always). Similarly, people indicated to what extent experiences this far caused people felt the **desire to give up on healthcare** (“Taking all these experiences into account did you ever feel like giving up trying to get your symptoms explained (or diagnosed)?”) from (never, to always).

Data analysis

Data were examined to determine suitability for analyses (data screening, missing data analyses). Data screening included recoding data into numeric values where these entered as textual data (e.g. ‘8 weeks’, was converted to ‘8’). ‘Number of doctor visits’ was difficult for people to estimate because of the high number of appointments many women had experienced. For number of doctor visits the decisions were: if people indicated an amount over a specific number (e.g., 10+ visits) the value entered was the minimum (i.e., 10); if a range was provided (i.e., 30 to 40 visits) the mid-range was entered (35). People who indicated inadvertent discovery of endometriosis (e.g., upon examination for appendicitis, cervical cancer) were given a code of zero to recode this possibility. However, because the latter biases the number of visits downward, these participants were indicated separately. If people indicated the number of years of appointments instead of number of visits they were coded as ‘missing’. If people indicated ‘too many to count’ they were coded separately. One outlier (480 visits) was excluded from analysis. Note that the question asked ‘number of visits made to your doctor prior to diagnosis’

II. Qualitative Workshop

The Endometriosis workshop followed the recommended format (see Gameiro et al for full details). The workshop started with an ice-breaking activity to allow participants and researchers to introduce themselves. Issues about the group format, respect and confidentiality were discussed and agreed. Participants were introduced to the basics of drawing (how to draw things and people) and guided through some simple drawing exercises (e.g., transforming plain circles into an object of their choice, a face expressing an emotion, etc). Participants were encouraged to use colour as they thought appropriate. The participants were then introduced to the concept of visual metaphor, defined simply “...as the use of something visible to show something that is invisible” (Gameiro et al) with examples provided. Three drawing activities were used to collect data. First, women were asked to **draw a self-portrait** and share it with the group. Second, participants were asked to **draw their symptom experiences**: ‘If endometriosis was an object, creature or animal or weather, what would it be?’). Third, participants were asked to **draw their experiences of healthcare**: ‘How would you draw your relationship with medical staff?’ After each exercise participants shared their artwork with the group, followed by group discussion of emerging views. There were three drawing tables (3 to 4 participants each) and simultaneous audio recorders at each table captured table and group discussions. Three researchers facilitated the workshop (rationale for research, DrawingOut Health methodology, drawing techniques) and took notes at each table to support the transcription process. The audio recordings were transcribed verbatim for analysis by an independent

Data Analysis

Boivin and Kingwell undertook thematic qualitative analysis according to well-established methodology (see Braun and Clarke (2006). As per other DrawingOut Health workshops this was the preferred analytic approach because data collection was concluded at the time of the analysis, the data consisted of transcribed material, and there was no strong theoretical perspective driving the data description (Howitt, 2010). However, the Health Belief Model was only applied the final stages of data analysis, to interpret links between broad themes identified.

The aim of the analysis was to derive a thorough description of endometriosis as emerged during the workshop. We used a bottom-up (inductive) coding process that derives, in its first step, a set of codes that closely fits transcribed data with minimal inference-making (Braun and Clarke, 2006: 83). Briefly, the analytic process involved familiarisation with the full workshop transcript, assigning textual descriptors (i.e., codes) to all textual passages that contained relevant content, discussing the descriptors until no new codes emerged, grouping thematically related codes into more abstract higher-order clusters (i.e., categories) through similar inductive coding, and, finally, identifying overarching ideas (i.e., broad themes) from the categories and their relation to each other that expressed key findings of the data collection (Braun and Clarke, 2006: 82). The coding (codes, categories, broad themes) was discussed with the other workshop facilitators. Of the four researchers, none had experience of endometriosis, two (JB, SG) are reproductive health academics and psychologists with in-depth knowledge of the disease, one (HK) was a medical student with 2 years of clinical patient experience, and one (LER) an academic with expertise in visual and multimodal forms of communication.

In the final stage of analysis (to be completed), a booklet produced from the coding will be presented to the workshop participants, who will be asked to comment on the degree to which they felt the booklet represented their views and experiences. After agreeing themes a graphic designer will be commissioned to produce a booklet reflecting the broad themes using participant drawings and representative quotes. The booklet will be used to convey participants' views of endometriosis and provide a vehicle to share the results of the study. At the end of the booklet a brief anonymised description of the participants and a toolkit with infertility-related information and support for readers will be shown. Only minor changes to participants' artwork (e.g., adding colour) will be carried out to enhance coherence.