Validity of self-reported endometriosis and endometriosis-related questions in a Swedish female twin cohort

Rama Saha, M.D., Lena Marions, Ph.D., and Per Tornvall, Ph.D.

Department of Clinical Science and Education Södersjukhuset, Karolinska Institutet, Stockholm, Sweden

Objective: To examine the validity of self-reported endometriosis and to improve the reliability of questionnaires by including endometriosis-related questions.
Design: Analysis of survey questionnaire data.
Setting: Cross-sectional study.
Patient(s): Cohort of 26, 898 female twins aged 20–60 years at interview, who participated in either of two surveys (1998–2002 or 2005–2006).
Intervention(s): None.
Main Outcome Measure(s): Endometriosis diagnosis in the Swedish National Inpatient Registry (IPR).
Result(s): The self-reported endometriosis diagnoses and endometriosis-related questions from a nationwide population-based twin registry were linked with the IPR. Fairly good agreement was found between the self-reported and IPR data on endometriosis. The receiver operating characteristics (ROC) curves showed fairly good predictive ability of self-reported endometriosis to have a confirmed endometriosis diagnosis in the IPR with an area under the curve (AUC) 0.79 (95% confidence interval [CI], 0.77–0.81). Further, the predictive ability increased to AUC 0.89 (95% CI, 0.88–0.90) when there was additional information about infertility and age.
Conclusion(s): Our results indicate that self-reported data on endometriosis are moderately accurate and may be useful in studies when register data are not available. (Fertil Steril® 2017;107:174–8. ©2016 by American Society for Reproductive Medicine.)
Key Words: Endometriosis, in-patient register, questionnaire, self-report

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Epidemiologic studies are often performed either by using national registers with health-care–based diagnoses or by questionnaires that include self-reported diagnoses. Several investigators have reported excellent agreement between self-reported endometriosis and medical records (1–3). Dysmenorrhea, chronic pelvic pain (CPP), deep dyspareunia, cyclical intestinal complaints, fatigue/weariness, and infertility continue to be the leading symptoms and signs of endometriosis (4–7). Dysmenorrhea was the chief complaint, reported by 62% of women with mainly peritoneal endometriosis in a Brazilian study (7). In the same study, the prevalence of CPP was 57%, deep dyspareunia 55%, cyclic intestinal complaints 48%, and infertility 40%. One review by Guo and Wang (8) that included 27 publications based on estimation of prevalence of surgically confirmed endometriosis showed that the average prevalence of endometriosis in women with self-reported CPP was 28.7% (95% CI, 27.0, 30.4).

It was reported by Meisinger et al. (9) that the postal questionnaire method seems to be a useful method to identify incident nonfatal acute myocardial infarction cases treated in a hospital in a epidemiologic cohort study. One very recent Norwegian study reported good concordance between self-reported hypertension and/or proteinuria during previous pregnancies and actual clinical findings among the cases (10). De Boer et al.

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Reprint requests: Rama Saha, M.D., Department of Clinical Science and Education Södersjukhuset, Karolinska Institutet, Sjukhusbacken 10, Stockholm 118 83, Sweden (E-mail: rama.saha@ki.se).

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(11) in one study on validity of self-reported causes of subfertility with medical records reported that tubal and male subfertility were highly accurate but other causes had low to moderate accuracy.

Twin studies often use self-reported data when assessing reasons for individual differences in terms of genetic and environmental influences of a trait or disease. The validity of self-reported hyperthyroidism and hypothyroidism when comparing self-reported questionnaire data in twins with medical records was shown to be unsatisfactorily low (12), but higher rates of agreement were found for osteoarthritis based on self-reported data in twins with clinical and radiographic classification criteria for osteoarthritis (13). One recent study compared self-reported social security data in a Swedish twin cohort with national insurance registry data to assess the validity of the self-reported data. That study showed that self-reported disability pension data may be very useful in studies when registry information is not available, but registry data was preferred, especially for long-term sickness absences (14).

To our knowledge, no reports are available on the validity and reliability of self-reported endometriosis and endometriosis-related questions using national inpatient registers. Therefore, we examined the validity of self-reported endometriosis for improving the reliability of questionnaires by including endometriosis-related questions in a sample of female twins aged 20 to 60 years in the Swedish Twin Registry (STR) who had endometriosis diagnoses in the Swedish National Inpatient Registry (IPR).

MATERIALS AND METHODS

Data Sources

We obtained data from the nationwide population-based STR and the IPR. The STR is the largest twin registry in the world and includes all twins born in Sweden since 1886, constituting more than 194,000 twins and more than 75,000 twin pairs (15). We used data from two previous cross sectional surveys conducted at STR: the Screening Across the Lifespan Twin (SALT) Study (16) performed from 1998 to 2002 via telephone interviews among twins born between 1926 and 1958 (aged >40 years at the time of data collection), and the Swedish Twin Study of Adults' Genes and Environments (STAGE) (17) performed from 2005 to 2006 by way of a Web-based questionnaire among twins born between 1959 and 1985 (aged 20–40 years at the time of data collection). The IPR includes patients treated through inpatient care in public hospitals in Sweden. This register was initiated in 1964, covered 60% of the Swedish population in 1969, 85% in 1983, and close to 100% since 1987. The discharge diagnoses of endometriosis in the IPR are coded according to the International Classification of Diseases 8, 9, and 10 (ICD 8–10), which are listed in Supplemental Table 1 (available online).

Linkage between the registers was made by use of Personal Identity Numbers (PINs), which are unique identifiers specific to each individual in Sweden (18). One recent study from Sweden compared diagnoses between medical records and diagnoses in the IPR and reported that 98% had a correct endometriosis diagnosis in the IPR, and 99.5% had an

TABLE 1

Response of self-reported endometriosis and endometriosis-related questions among women aged 20–60 years in Swedish Twin Registry with endometriosis diagnosis in the Swedish National Inpatient Registry.

	Endometriosis in IPR		
Questionnaire in STR	**Yes, n (%)**	**No, n (%)**	**Missing values, n (%)**
Age at interview, y			
20–30	8 (1.33)	4,835 (18.4)	0 (0)
31–40	52 (8.6)	4,826 (18.4)	0 (0)
41–50	213 (35.4)	7,962 (30.28)	0 (0)
51–60	329 (54.65)	8,673 (33.0)	0 (0)
Self-reported endometriosis			
Yes	372/602 (61.8)	796/26,296 (3.0)	0 (0)
No	230/602 (38.2)	25,500/26,296 (97.0)	0 (0)
Severe dysmenorrhea			
Yes	123/602 (20.4)	5,680/26,296 (21.6)	
No	91/602 (15.1)	12,990/26,296 (49.4)	8,014 (30.5)
Chronic pelvic pain			
Yes	53/602 (8.8)	2,029/26,296 (7.7)	8,014 (30.5)
No	161/602 (26.7)	16,641/26,296 (63.3)	
Dyspareunia			
Yes	34/602 (5.7)	752/26,296 (2.9)	8,014 (30.5)
No	180/602 (3.0)	17,918/26,296 (68.1)	
Infertility			
Yes	167/602 (27.7)	1,900/26,296 (7.2)	0 (0)
No	435/602 (72.3)	24,392/26,296 (92.8)	0 (0)
Oral pill as contraceptive			
Yes	99/602 (16.5)	5,297/26,296 (20.1)	0 (0)
No	503/602 (83.6)	20,999/26,296 (79.9)	0 (0)

Note: IPR = Swedish National Inpatient Registry; STR = Swedish Twin Registry.

Saha. Validity of self-reported endometriosis. Fertil Steril 2016.

TABLE 2

Measures of agreement between self-reported and national register data on endometriosis in a Swedish twin cohort.

Questions in STR	Sensitivity (%)	Specificity (%)	ROC area
Self-reported endometriosis	61.8	97.0	0.79
Severe dysmenorrhea	57.5	69.6	0.64
Chronic pelvic pain	24.8	89.1	0.57
Dyspareunia	15.9	96.0	0.56
Infertility	27.7	92.8	0.60
Oral pill as contraceptive	16.4	79.9	0.48

Note: ROC = receiver operating characteristics; STR = Swedish Twin Registry.

Saha. Validity of self-reported endometriosis. Fertil Steril 2016.

endometriosis diagnosis confirmed by surgery (19). In 1997 visits to day surgery clinics began to be included in the IPR; since 2001 outpatient visits have been registered in the IPR.

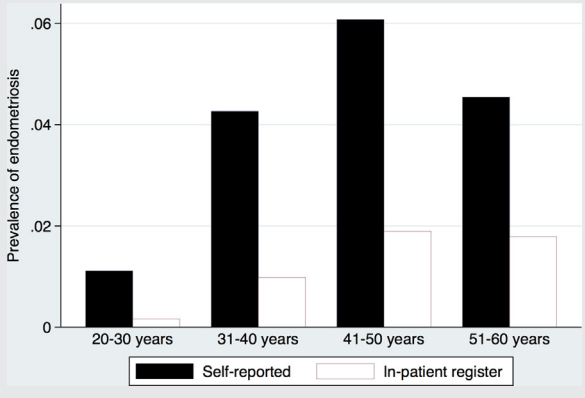
Data Collection and Methods of Analyses

Participants. The study group comprised the SALT and STAGE cohorts, including a total of 26,898 female twins aged 20–60 years at the time of the interview. We excluded women who were more than 60 years of age at the time of interview (in SALT); the IPR had attained close to 100% coverage since 1987, so the women older than 60 years might have missed being registered in the IPR.

In the questionnaire, the women were asked "Have you ever been diagnosed with endometriosis, also called chocolate cysts?" The answer was yes for 1,168 women ($\leq$60 years). By linking the STR and IPR at survey (interview time) we could find 602 cases of endometriosis in the IPR. Women were also asked endometriosis-related questions, which are listed in Supplemental Table 2 (available online).

We calculated descriptive information of endometriosis-related questions for participants with and without endometriosis as absolute numbers and percentages. We determined the validity measures sensitivity and specificity.

FIGURE 1



Prevalence of endometriosis according to age categories.

Saha. Validity of self-reported endometriosis. Fertil Steril 2016.

Logistic regression analysis was used to determine which variables were independently associated with overall agreement regarding the endometriosis diagnosis reported in the questionnaires and in the IPR. The variables of interest were age at interview (20–30, 31–40, 41–50, and 51–60 years), endometriosis, and infertility. Other variables were excluded for this analysis because all these variables had missing values for about 8,000 individuals. We plotted the sensitivity and specificity on a receiver operating characteristics (ROC) curve to determine the prediction of an endometriosis diagnosis in the IPR. An area under the curve (AUC) equal to 0.50 signals random prediction, an AUC of 0.60–0.70 indicates poor validity, 0.70–0.80 is fair, 0.80–0.90 is good, and >0.9 shows an excellent validity (20). All analyses were processed with Stata IC 12 software (StataCorp).

The study was reviewed and approved by the Regional Ethics Committee in Stockholm, Sweden (diary number 2009/1676–31/2).

RESULTS

Table 1 presents the responses of self-reported endometriosis and endometriosis-related questionnaires in the STR with endometriosis diagnosis in the IPR. There were no nonresponses for endometriosis, age, infertility, or oral pill as a contraceptive. The nonresponse was about 30% for severe dysmenorrhea, pelvic pain, and dyspareunia.

Table 2 presents the sensitivity and specificity of self-reported endometriosis and endometriosis-related questionnaires in STR for endometriosis diagnoses in IPR. Results for all variables show a high specificity except for severe dysmenorrhea while sensitivity was low.

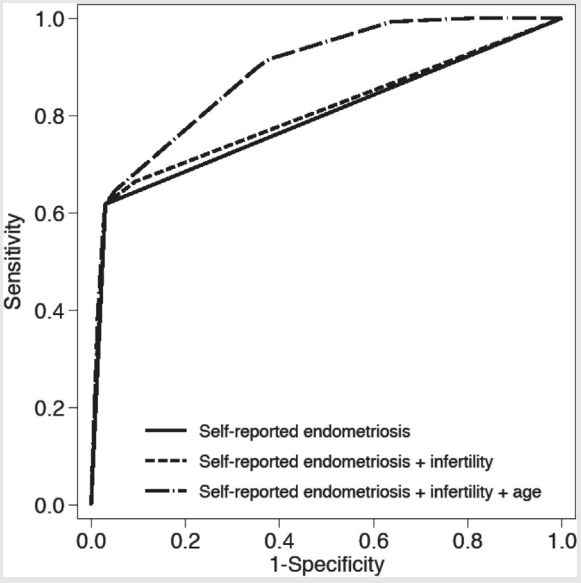
Figure 1 presents the prevalence of endometriosis according to age categories at interview. It shows that the prevalence of endometriosis is directly associated with age, with no further increase after 40 years in the self-reporting women and after 50 years in the IPR.

Figure 2 presents ROC curves illustrating the fairly good predictive ability of self-reported endometriosis to have a confirmed endometriosis diagnosis in the IPR with an AUC 0.79 (95% confidence interval [CI], 0.77–0.81). Further, the predictive ability increases to AUC 0.80 (95% CI, 0.78–0.82) when there was additional information about infertility, and the predictive ability increases to AUC 0.89 (95% CI, 0.88–0.90) when there was additional information about age and infertility.

DISCUSSION

To our knowledge, this is the first study on the validity and reliability of self-reported data on endometriosis and endometriosis-related questions using the IPR. Fairly good agreement was found between the self-reports and the IPR data on endometriosis. Further, the predictive ability of self-reported endometriosis increases when there was additional information about age and infertility. We found high specificity of self-reported endometriosis in the present study, showing that the women who answered no could correctly classify themselves based on self-reports, which is in line with previous findings (14).

FIGURE 2



Receiver operating characteristics for prediction of endometriosis diagnosis in the inpatient register.

Saha. Validity of self-reported endometriosis. Fertil Steril 2016.

Previous studies have evaluated the agreement between self-reported data (9, 14) and register data on other questions but not on endometriosis. However, previous studies have compared self-reported endometriosis data with medical records with good agreement (1–3). One recent study compared self-reported social security data in a Swedish twin cohort with national insurance registry data to assess the validity of self-reported data. The study showed that self-reported disability pension data may be very useful in studies when registry information is not available, with a sensitivity of 70% and specificity of 99%; however, registry data is preferred, especially for long-term sickness absence due to low sensitivity of 45% (14). Another study assessed the agreement between myocardial infarction reported in a postal questionnaire with data from a register in a representative sample of German men and women (n = 9,176) aged 25 to 74 years and reported good agreement, where sensitivity was as high as 98.0% and specificity 99.3%. They concluded that postal questionnaire was a useful method to identify hospitalizations for incident nonfatal acute myocardial infarction cases in epidemiologic cohort studies (9).

De Boer et al. (11) in one study on the validity of self-reported causes of subfertility assessed the accuracy of causes of subfertility as reported by women in a self-administered questionnaire in comparison with medical record information in a nationwide cohort study of women receiving in vitro fertilization treatment in the Netherlands. They observed that the sensitivity, positive predictive value, and agreement as expressed by kappa were excellent for tubal (84%, 91%, and 0.79, respectively) and male subfertility (87%, 78%, and 0.71, respectively), but other causes were low to moderately accurate. In a Norwegian retrospective case-control study with 200 randomly selected cases and 200 controls on the validation of self-reported information about hypertensive disorders of pregnancy, Falkengård et al. (10) assessed the actual clinical findings among the cases by using medical records. They reported that self-reported information on hypertensive disorders of pregnancy has appropriate validity to be used for epidemiologic research.

All four studies mentioned here concluded that self-reported data may be very useful in studies when registry information is not available. Our study is in line with those studies mentioned earlier.

Among those who reported severe dysmenorrhea in the current study, 20.4% had an endometriosis diagnosis in the IPR, while dysmenorrhea was reported as a chief complaint by 62% of women with mainly peritoneal endometriosis confirmed by medical records in a Brazilian study (7). Of women who responded positively to pelvic pain question in the STR, 8.8% had an endometriosis diagnosis in the IPR. However, one previous review based on 27 published studies (including only small samples) on the prevalence of endometriosis in CPP, reported that the average prevalence of endometriosis in women with CPP was 28.7% (8). Studies have suggested that 25% to 50% of infertile women have endometriosis and that 30% to 50% of women with endometriosis are infertile (1, 21). The present study figure that 27.7% of infertile women had an endometriosis diagnosis in the IPR is consistent with previous studies. Women with self-reported dyspareunia had an endometriosis diagnosis in 5.7% of cases. A recent Brazilian study reported that deep dyspareunia was associated with endometriosis in 55% of cases (7).

The current study showed the fairly good predictive ability of self-reported endometriosis to have an endometriosis diagnosis in the IPR. The predictive ability of self-reported endometriosis increased from 79% to 80% when there was additional information about infertility and increased from 79% to 89% when there was additional information about infertility and age. Women are usually affected by endometriosis at a younger age. Age is not assumed to be causally associated with endometriosis, but it can take years to have the endometriosis diagnosis confirmed; thus older women with endometriosis are more likely to have their disease registered in the IPR as compared with younger women.

Strengths and Limitations

The main strength of our study was the large cohorts from the population-based STR. Studies suggest that the results of twin studies can be generalized to singleton populations (3, 22, 23). These studies also rely on the accurateness of self-reported data not being influenced by twin status (zygosity) in itself; that is, whether the twin is identical (monozygotic) or fraternal (dizygotic) should not influence the way of reporting the presence or absence of health symptoms, diseases, or other factors in surveys. Thus, the STR is representative for the Swedish general population.

Our study is also subject to limitations. Dysmenorrhea, dyspareunia, and CPP had a 70% response rate in STR, and thus the true prevalence of endometriosis in the IPR

associated with these symptoms was not possible to estimate. Another limitation is the retrospective nature of the study including only subjective information regarding endometriosis which made it impossible to differentiate between different forms of the disease. Use of inpatient register data could lead to an underestimation of the prevalence of endometriosis in this population because visits to day surgery clinics only began to be included in the IPR in 1997 but since 2001 all outpatient visits are registered in the IPR. Further, in the present study, the question on "severe dysmenorrhea" might not have been the very best one, because "severe" might be interpreted in many different ways by the respondents. The present study was also subject to recall bias due to different time spans. Women were asked about symptoms and signs many years after they had first experienced them. Although we tried to limit recall bias through the use of a highly structured questionnaire, there is perhaps a possibility that responses have been differentially recalled.

CONCLUSION

Our results indicate that self-reported data on endometriosis is moderately accurate and may be useful in studies when register data are not available. Further, additional information about age and infertility could improve the results. It is advised that surveys of endometriosis should include validated instruments or, in the absence of such, clearly phrased questions in interviews or by questionnaire.

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SUPPLEMENTAL TABLE 1

International Classification of Diseases (ICD) of endometriosis.

Revision 8, 1965	
625.30	Endometriosis of ovary
625.31	Endometriosis of pelvic peritoneum
625.32	Endometriosis in retro-cervical and retro-uterine area
625.33	Endometriosis of uterus (Adenomyosis)
625.38	Other endometriosis
625.39	Endometriosis with non-specific locations
Revision 9, 1987	
617A	Endometriosis of uterus (Adenomyosis)
617B	Endometriosis of ovary
617C	Endometriosis of fallopian tube
617D	Endometriosis of pelvic peritoneum
617E	Rectovaginal and vaginal endometriosis
617F	Endometriosis of intestine
617G	Endometriosis in cutaneous scar
617W	Endometriosis with other specific locations
617X	Endometriosis with non-specific locations
Revision 10, 1997	
N80.0	Endometriosis of uterus (Adenomyosis)
N80.1	Endometriosis of ovary
N80.1	Endometrioma
N80.2	Endometriosis of fallopian tube
N80.3	Endometriosis of pelvic peritoneum
N80.4	Endometriosis of rectovaginal septum and vagina
N80.5	Endometriosis of intestine
N80.6	Endometriosis in cutaneous scar
N80.8	Other endometriosis (Endometriosis of thorax)
N80.9	Endometriosis, unspecified

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SUPPLEMENTAL TABLE 2

Endometriosis-related questionnaire in the Swedish twin registry.

Have you ever been diagnosed with endometriosis, also called chocolate cysts?
Do/did you experience severe menstrual pain?
Do you take strong painkillers because of pain?
Have you been absent from work due to pain?
Do you take oral contraceptive pills because of menstrual pain?
Do you experience pelvic pain in between menstrual periods?
Do you experience painful intercourse?
Have you been investigated or treated for infertility?
Do you regularly use oral contraceptive pills as a contraceptive?

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