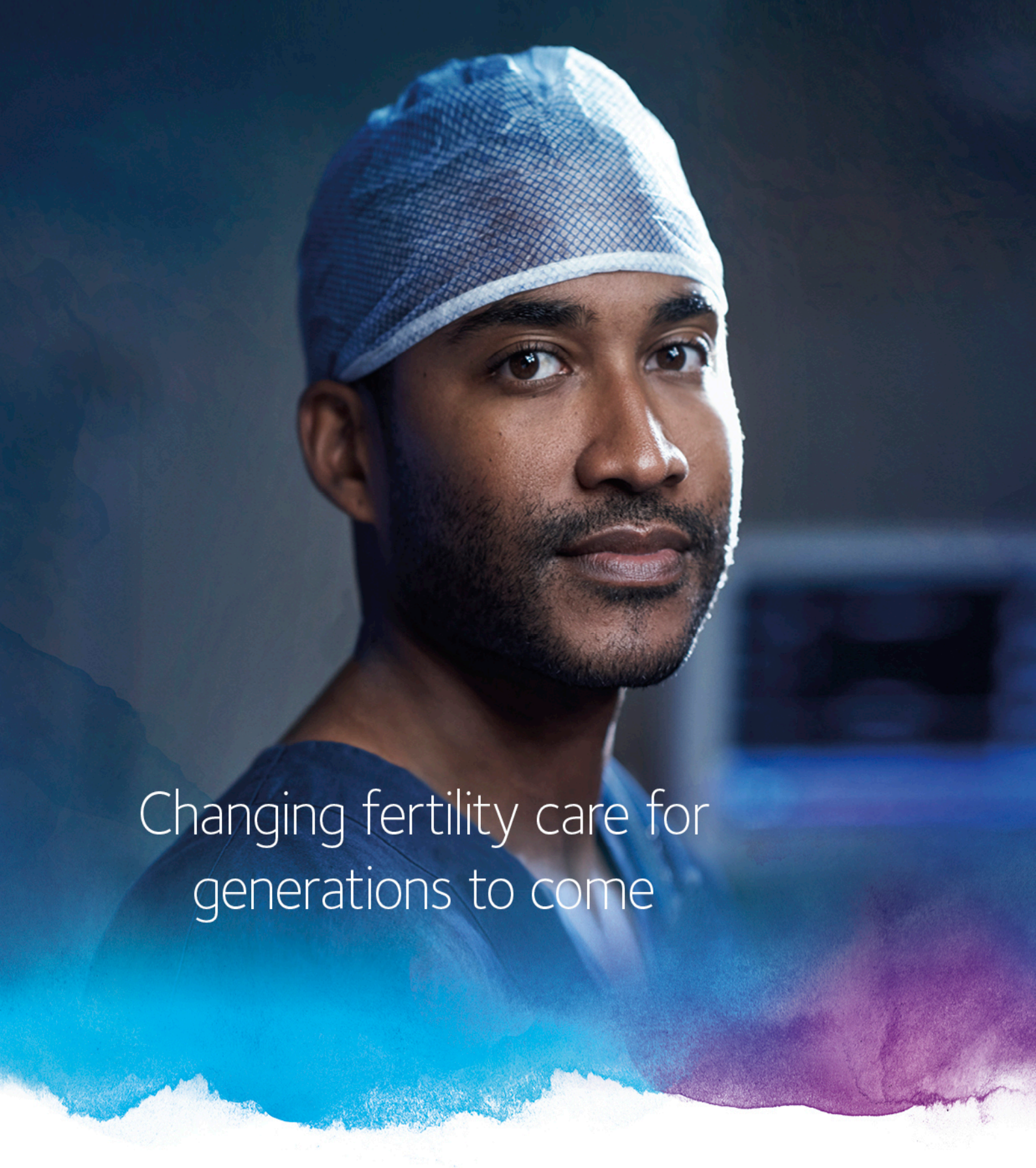

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BJOG An International Journal of Obstetrics and Gynaecology

Royal College of Obstetricians & Gynaecologists

DOI: 10.1111/1471-0528.16853
www.bjog.org

Research Article

Pregnancy denial: a complex symptom with life context as a trigger? A prospective case–control study*

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Received 26 December 2020; revised 20 May 2021;
Accepted 10 July 2021.

Objective To identify risk factors for a woman to experience pregnancy denial.

Design, setting and population A French multicentric prospective case–control study with 71 mother–infant dyads having experienced a pregnancy denial versus a control group of 71 dyads.

Methods Data were collected in the week after delivery using an observational leaflet and two psychiatric scales (MINI and QSSP).

Main outcome measures Statistically significant differences between the two groups regarding social, demographic, medical and psychiatric data.

Results Not being in a stable relationship (odds ratio [OR] 17.18, 95% CI 3.37–87.60]; $P < 0.0001$), not having a high school diploma (OR 1.11, 95% CI 1.04–1.38]; $P < 0.0001$) and having a psychiatric history (OR 6.33, 95% CI 1.62–24.76; $P = 0.0002$) were risk factors to experience pregnancy denial, whereas being older was a protective factor (OR 0.86, 95% CI 0.79–0.93; $P = 0.0054$) (logistic regression, Wald 95% CI). Other risk factors included late declarations of pregnancy history and past pregnancy denials (case $n = 7$, 9.7% versus 0% in controls; $P = 0.01$), past pregnancy denials in the family (case $n = 13$, 18% versus control $n = 4$, 5.6%; $P = 0.03$), and use of a contraceptive method (75% for cases versus 7% in control; $P < 0.0001$), primarily an oral contraceptive (75%).

Conclusion Family or personal history of pregnancy denial should be part of the systematic anamnesis during the first visit of a patient of child-bearing age. Further, our study points out that life context (young age, single status, socio-economic precarity, pill-based contraception) could be a trigger for pregnancy denial in certain women.

Keywords Age, education, life context, marital status, newborn, pregnancy denial, psychiatric background, risk factors.

Tweetable abstract Life context can be a trigger for pregnancy denial.

*A prospective case–control study with 71 mothers having experienced pregnancy denial in comparison with a control group of 71 mothers.

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Please cite this paper as: Delong H, Eutrope J, Thierry A, Sutter-Dallay A-L, Vulliez L, Gubler V, Saad Saint-Gilles S, Tessier E, Le Foll J, Viaux S, Apter G, Danion A, Auer J, Rolland A-C; the Pregnancy Denial Study Group. Pregnancy denial: a complex symptom with life context as a trigger?: A prospective case-control study. BJOG 2021; https://doi.org/10.1111/1471-0528.16853.

Introduction

Pregnancy denial was described by authors as early as the 17th century.[1,2] It is fascinating both for its still misunderstood physiopathology[3–5] and for its potential dramatic consequences (e.g. unexpected deliveries, rare neonaticides). Nonetheless, even among professionals, there is no consensus concerning its accurate definition. Pregnancy denial occurs when a mother-to-be is unaware of being pregnant for several months or even throughout the pregnancy. However, the threshold date from when the pregnancy is considered to be denied if unacknowledged, divides professionals: beyond 14 weeks of amenorrhoea,[6,7] 21 weeks of amenorrhoea,[8] 20 weeks of pregnancy[9] or the end of the third trimester.[10] There is also ambiguity between pregnancy denial, which is unconscious, and pregnancy dissimulation, which is, on the contrary, conscious. Mainly due to this imprecision, pregnancy denial is not yet included as a pathology in Diagnostic and Statistical Manuals-5 V or International Classification of Diseases-10 and remains poorly understood by the medical profession: 'the lack of attention to the phenomenon of pregnancy denial mirrors the silent stance of these patients' (Friedman et al., 2007).[10]

Yet, the prevalence of pregnancy denial has been estimated to be 1 case in 475 births,[9] making it far more common than a rare disease (below 1/10 000 cases). Moreover, the consequences can be serious for the mother and the newborn, especially due to a late or non-existent obstetrical follow-up or continuation of teratogen habits (tobacco, alcohol, etc.)[8,11] or to less obvious reasons such as potential psychological distress for the mother or impaired interactions between mother and child.[12–14]

Since the 1990s, a few epidemiological studies (Appendix S1) have tried to better document pregnancy denial. They have suggested that women experiencing pregnancy denial were mostly middle-aged,[10,15] more often unemployed[15–17] and single[10,15,16] compared with the general population, and without any psychiatric history.[10,15,16] However, those studies dealt with limitations that were either statistical (small samples,[8,17] no statistical analysis[8]) or methodological (retrospective analysis,[10] no control group,[10] no structured psychiatric interview[10,15]).

To further improve our knowledge of pregnancy denial, we conducted a case–control prospective study with 142 French mother–infant dyads (71 women having experienced pregnancy denial and 71 having not), from 2013 to 2019.[18] In this article, we analysed collected socio-demographic, medical and psychiatric data. Our objective was to identify risk factors for a woman to experience pregnancy denial.

Methods

Study setting and context

This study was based on data from a national multicentre prospective case–control study with 13 French university maternity wards (Antony Ile-de-France, Besançon, Bordeaux, Nancy, Paris Bichat, Paris La Pitié Salpêtrière, Reims, Strasbourg, Toulouse, Amiens, Troyes, Aubagne, and Lille) entitled 'Attachment and Pregnancy Denial' conducted from 2013 to 2019.[18] The study compared two groups of 71 mother–infant dyads each: a 'case' group with maternal pregnancy denials and a 'control' group without pregnancy denials. The study was registered in the European registry (EudraCT 2011-A01498-33) and in clinicaltrials.gov (NCT02867579). It was supported by grants from the Hospital Research Program of the French Ministry of Health (PHRC 2011). The funders had no role in the study design, data collection and analysis, decision to publish, or preparation of the manuscript.

Patient involvement

Regarding the inclusion criteria, in the case group, the dyads comprised a woman with pregnancy denial and her infant. Pregnancy denial is defined as a pregnancy announcement after 20 weeks of gestation and a lack of objective perceptions of pregnancy by the woman. In the control group, the dyads comprised a woman without pregnancy denial and her infant. Cases and controls were matched in real-time on primiparous and non-primiparous women and on premature (birth before gestation week 37) and non-premature infants.

The exclusion criteria were as follows: age <18, intellectual disability, acute or chronic psychotic condition, not fluent in French, illegal administrative status, anonymous childbirth, medically assisted reproduction for this pregnancy, women with protective supervision, and newborn with a life-threatening prognosis or an organic malformation and/or genetic abnormality.

Assessment

The interviews were conducted at the maternity ward within a week after delivery. They were led by a psychologist or psychiatrist with training and experience in perinatal period. The consent of the mother was necessary in all cases, as well as that of the father if he had legal co-responsibility for the child.

Data were collected using an observational leaflet and two specific psychiatric scales:

1 The observational leaflet was specially designed for this research. Data collected from mothers included administrative and socio-demographic information, clinical information, medical background, history of pregnancy, denials in the family, data on recent pregnancy, and classical birth information of the child (birth date, term, weight, etc.).
2 Psychiatric questionnaires.

The Mini International Neuropsychiatric Interview (MINI) is a structured interview that explores the main psychiatric disorders of the axis I of the 10th revision of the International Classification of Diseases (ICD-10). With an administration time of approximately 15 minutes, the scale is divided into 16 modules, each corresponding to a diagnosis category. The MINI notably allows the detection of a major depressive disorder and points out potential psychiatric comorbidities.

The Perceived Social Support Questionnaire (PSSQ) allows a direct assessment of perceived social support over the past few months. This is a self-assessment scale.

Sample size calculation

Given the average number of births per year and by centre (2500 on average), number of participating centres ($n = 13$), frequency of pregnancy denial (1/500 child births), duration of the inclusion phase (48 months) and expected participation acceptance rate (25%), 70 women with pregnancy denial were planned to be included in this study, in total, 140 mother–infant dyads. Eventually, 142 dyads were successfully included (71 in the case group and 71 in the control group).

Statistical analysis

Qualitative variables were described through numbers and percentages and compared through chi-square or Fisher exact tests. Quantitative variables were described through means and standard deviations or medians and extremes and compared through univariate analysis using Student or Wilcoxon–Mann–Whitney tests (according to the conditions of application of tests). For all these analyses, the significance level was set at 0.05. A logistic regression was calculated from variables with a significance level <5% on univariate analysis. Statistical analysis was performed using SAS version 9.4 (SAS Inc., Cary, NC, USA).

Results

Administrative and socio-demographic data of mothers

Case women were younger than control women (24 versus 30 years old; $P < 0.0001$) (Table 1). They were also more frequently single (37% versus 3%; $P < 0.0001$) and, if with a partner, they were less often with the child's father at the time of pregnancy discovery than were control mothers (89% versus 99%; $P < 0.0001$).

Table 1. Administrative and socio-demographic data of mothers

Variables	Case group ($n = 71$)	Control group ($n = 71$)	*P*-value
Age	24 [18;40]*	30 [18;42]*	< 0.0001
Location (%)			0.58
Rural	27	32	
City	73	68	
Marital status at the discovery of the pregnancy (%)			< 0.0001
Single	37	3	
In a couple	63	97	
If in a couple			0.03
With the child's father	89	99	
With another person	11	1	
High school diploma (%)			< 0.0001
Yes	42	87	
No	58	13	
Activity (%)			0.0003
Yes	57	85	
No	43	15	

*Median [min;max].

Case women had a high school diploma significantly less often (42% versus 87%; $P < 0.0001$) and were more often out of a job or a training at the time of the interview (43% versus 15%; $P = 0.0003$).

When engaged in work (job or training/studies), case women were overrepresented among employees (56% versus 35% for control; $P = 0.04$) and students (27% versus 5%; $P = 0.002$), whereas control women were predominantly executives (43% of control women versus 2% of cases; $P < 0.0001$).

Clinical data of mothers

There was no significant difference between case women and control women concerning their body mass index before pregnancy, gestational age, parity, obstetrical history and medico-chirurgical background (Table 2).

Case women declared a psychiatric background (diagnosis, psychiatric care or psychological counselling, hospitalisation, long-term prescription of psychotropics) significantly more often ($P = 0.02$) than control women did.

The MINI questionnaire was used to investigate this background. A significant difference was found between the two groups with respect to depressive disorders (background of major depressive disorder or persistent

Table 2. Clinical data of mothers

Variables	Case group (n = 71)	Control group (n = 71)	P-value
Weight before pregnancy (kg)	62 [38;115]*	62 [45;100]*	0.56
Height (cm)	164 ± 7.14**	164.6 ± 6.60**	0.63
BMI	23.7 [16.6;43.3]*	22.8 [17.1;36.7]*	0.36
Gestity	1 [0;7]*	1 [0.7]*	0.83
Parity	1 [0;6]*	1 [1;7]*	0.92
Primiparity (%)	66.2	68.6	0.15
Obstetrical history (%)	38.9	40.9	0.87
Among which,			
Miscarriage	28.6	31	1.0
Therapeutic abortion	3.6	3.5	1.0
Legal abortion	60.7	48.3	0.43
Assisted procreative techniques	0	3.45	1.0
Pre-term birth	10.7	10.3	1.0
Threatened premature labor	10.7	10.3	1.0
Hospitalisation linked to the pregnancy	21.4	10.3	0.30
Caesarean section	25	6.9	0.08
Psychiatric or psychological follow-ups during past pregnancies (%)	2.3	8.7	0.16
Medical background %)			
Chronic diseases	16.7	22.5	0.4
Long-term treatments	9.7	16.9	0.23
>1 week hospitalisation in a surgery unit	19.4	15.5	0.66
Eating disorders history (%)	8.3	2.8	0.28
Consumption prior to recent pregnancy (%)			
Alcohol	45.8	38	0.4
Tobacco	62.5	31.4	0.0002
Drugs	13.9	4.2	0.04
Psychiatric history (%)	23.6	8.5	0.02
Psychiatric diseases	13.9	7	0.28
Psychiatric follow-up	9.7	7	0.76
Psychological follow-up	12.5	15.5	0.64
Hospitalisation in a psychiatric unit	5.6	1.4	0.37
Long-term psychoactive treatments	6.9	4.2	0.72
Past late declaration of pregnancy (>20 wk of amenorrhea) (%)	9.72	0	0.01
Past pregnancy denials (%)	9.72	0	0.01
Past family pregnancy denials (%)	18.1	5.6	0.04
Psychiatric diseases in the family	22.2	19.7	0.84

*Median [min;max].
**Mean ± SD.

depressive disorder) (18.1% of case women versus 7.0% of control women; P = 0.047).

There was no significant difference between the two groups with respect to a history of eating disorders (P = 0.28).

Further, case women declared more active smoking of tobacco (63% versus 31%; P = 0.0002) and consumption of drugs before pregnancy (13.9% versus 4.2%; P = 0.04), but not more alcohol consumption (P = 0.40).

A history of late declarations of pregnancy (after 20 weeks of amenorrhoea) and past pregnancy denials was only found in the case group (n = 7, 9.7% versus 0% in the control group; P = 0.01), and past pregnancy denials occurred more often in the families of case women than in those of control women (n = 13, 18% versus n = 4, 5.6%; P = 0.03).

Data on recent pregnancy

Regarding the *type of pregnancy denial*, among case women, 19% (n = 10) had total pregnancy denial, that is, they discovered their pregnancy when the labour began, whereas 81% (n = 43) discovered their pregnancy before this point (missing data correspond to fathers who refused to participate in the study and to transmit the child birthdate).

Regarding the *course of pregnancy*, most pregnancies in the case group occurred while using a contraceptive method (75% versus 7% for control women; $P < 0.0001$), which was mainly an oral contraceptive (for 75% of them).

During pregnancy, women in the case group experienced significantly less change in their weight (+8.5 versus +12 kg for control women; $P = 0.0007$) and breast size ($P < 0.0001$); for 86% of them, menstruations continued throughout the denial period (while only 4.3% of control women reported menstruations during pregnancy; $P < 0.0001$). Case mothers also perceived less fetal movement (69% versus 98.6%; $P < 0.0001$). This obviously contributed to the lack of pregnancy awareness among case mothers.

Regarding *support*, women from the case group reported being less satisfied than control women by the support received from the child's father (69.2% versus 91.4%; $P = 0.001$), which is consistent with their single status or status of not being in a couple status with the father for 44% of case women versus 4% for controls (see Table 1).

In the case group, women received significantly more specific support during pregnancy (50.0% versus 21.1%; $P = 0.0003$), particularly from child and maternal protection centres and social workers. Almost all of them received specific support at the maternity unit (97.2% versus 11.3% for the control group; $P < 0.0001$), mainly via psychiatric or psychological interviews and child and maternal protection centres.

Regarding the *risk factors of pregnancy denial*, a logistic regression with a 95% confidence interval showed that not being in a stable relationship, not having a high school diploma and having a psychiatric history were risk factors for pregnancy denial, whereas being older was a protective factor (Table 3). For this regression, we only selected explanatory variables that had a significant level <0.05: age, marital status at the discovery of the pregnancy, high school diploma, consumption of alcohol, tobacco and drugs prior to recent pregnancy, psychiatric background, past late declaration of pregnancy (>20 weeks of amenorrhoea), past pregnancy denials and past family pregnancy denials.

Data of children

Data were gathered from 126 newborns, 60 in the case group (85%) and 66 in the control group (93%) (the missing data can be attributed to fathers who had legal authority and refused to transmit data).

There were no significant differences in sex, weight, cranial perimeter, neonatal reanimation, feeding mode and medical condition of the newborns. However, among the case group, the median pregnancy term (38 weeks) and median height of newborns (49 cm) were lower than those of the control group (39 weeks and 50.5 cm; $P = 0.01$, respectively).

Table 3. Logistic regression

Variables	OR (Wald 95% CI)	*P*-value
Not in a couple	17.18 (3.37–87.60)	< 0.0001
No high school diploma	1.11 (1.04–1.38)	< 0.0001
Psychiatric history (%)	6.33 (1.62–24. 76)	0.0002
Age	0.86 (0.79–0.93)	0.0054

Discussion

Main findings

The results showed that women who experienced pregnancy denial were younger, less educated, in a more precarious working situation, and more often single than control women were. They also reported a depressive background more often. In this sense, the logistic regression allowed us to conclude that not being in a stable relationship, not having a high school diploma, and having a psychiatric history are risk factors for pregnancy denial, whereas being older is a protective factor.

Strengths and limitations

To the best of our knowledge, this is the first published case–control prospective study on pregnancy denial dealing of such a sample size. The data collected were mainly factual (socio-demographic) or gathered through validated psychiatric questionnaires (MINI and QSSP).

The main limitation is that the diagnosis of pregnancy denial is by nature retrospective and based on mothers' declarations. However, to minimise some potential classifications biases, interviews were led only by skilled perinatal psychiatrists or psychologists.

Interpretation in the light of other studies (Appendix S1)

In our study, in line with Brezinka et al.,[8] classical signs of pregnancy, such as amenorrhoea, weight gain or increased breast size, are usually missing in cases of pregnancy denial.

Among physiopathological explanations of this phenomenon, a deficit in HCG or progesterone during pregnancy was assumed.[4,5,19] Other authors suggested that higher perceptive thresholds could result in women not perceiving changes in their bodies correctly.[20]

In our study, we noted that most case women were using a contraceptive, which had possibly contributed to a false reassurance. Moreover, most were taking an oral contraceptive, which is likely to provoke spotting or irregular bleeding. Thus, bleeding linked to pregnancy could have been wrongly interpreted. Concerning weight gain, average BMI prior to pregnancy was normal for the case group and not

significantly different from the control group. Still, the former experienced less weight change. We can speculate about whether case women, being unaware of their pregnancy, were less prone to indulging themselves, and tried to limit the weight gain.

As another explanation for pregnancy denial, some authors hypothesised that case women might either be uneducated about pregnancy (very young or primiparous), simple-minded or have a psychiatric disorder.[10,21] Our results, like those of Wessel[15] and Beier,[22] showed that indeed case mothers were mostly primiparous, but in the same proportion as controls, and younger than controls (around 24); however, the fact that 75% of them used contraceptives proves they had some sense of pregnancy risk management. Furthermore, several studies proved that only a minority of case women were of diminished intelligence or had a severe psychiatric disorder.[15,17,22,23] However, to our knowledge, only Brezinka et al.'s protocol[8] included a systematic screening of psychiatric history through an interview with a psychiatrist. They found 30% of their sample had a history of mood or personality disorder. Other studies collected psychiatric history directly from medical records and sometimes with a limited scope (e.g. only schizophrenia and 'further severe psychiatric relevant symptoms' in the Wessel study[15]), which raises questions about potential undetected disorders. Our study is the first to screen psychiatric background with a validated tool (MINI). Finding that depressive disorders are overrepresented among cases leads us to wonder whether pregnancy denial is truly a pathology *per se* or rather a symptom of a preexisting or reappearing depressive disorder.

Furthermore, in our study, 37% of case women were single, versus 3% of control women. Our results are consistent with previous studies.[15,16] One hypothesis could be that some women, who are not in a steady affective relationship, prevent themselves from conceptualising and accepting the idea of a pregnancy. For those women, the nuclear family could represent, at least unconsciously, the ideal background to have a child; thus, if such conditions do not exist, a pregnancy will not be acceptable.

We also found that only 42% of case women had a high school diploma, and 57% had some form of job or were in training (56% employees, 27% students). Compared with the control group (87% with high school diploma and 85% with work, mainly executives), this shows a greater precarious situation among case women. Consistent results emerged from Chaulet and Wessel's studies.[16,15] In our study, case women were 5 years younger on average, explaining why they were more likely students. However, our results show that, despite the sampled women being over high school age, controlling for job/training status, fewer case women had a high school diploma and were executives. This suggests a lower education level among the case group. Thus, we can wonder whether a greater socio-economic uncertainty, here evidenced by a lower education level, can contribute to the impossibility among case women to apprehend consciously a pregnancy.

Lastly, it is interesting that a history of late declarations of pregnancy (after 20 weeks of amenorrhoea) and past pregnancy denials were only found in the case group and that past pregnancy denials occurred more often in the families of case women than in those of controls. Therefore, a past pregnancy denial, whether by the woman herself or someone in her family, seems to be a predisposing factor for a pregnancy denial as well.

Regarding the newborn, our findings differ from Wessel's study, wherein pregnancy denial had a significant effect on the newborn (term, birthweight <2500 g, intrauterine growth restriction, hospitalisation in a neonatal unit, and surgical delivery).[24] In our study, the only significant differences between cases and controls were median pregnancy term (38 versus 39 weeks for cases) and median height of newborns. We have not succeeded in explaining these differences. However, if confirmed by further studies, these findings should help to destigmatise and alleviate the potential guilt of mothers who have experienced pregnancy denial.

Conclusion

In conclusion, our study, like others before it, does not allow us to establish a standard profile of a 'pregnancy denier' that would allow for a systematic screening of a given population.[15,25] Nevertheless, it brings to light risk factors that can enhance vigilance.

The most actionable risk factor is the existence of a family or personal history of pregnancy denial. This question could legitimately and easily be part of the systematic anamnesis of the medical professionals to identify and inform women at risk of pregnancy denial.

Another risk factor could be a psychiatric background, especially depression. Even if the link between depression and pregnancy denial needs further investigation, in the meantime, in a context of pregnancy denial, we would recommend systematic and thorough screening for perinatal depression.

In addition to history of depressive symptoms, other identified risk factors (young age, single status, socio-economic precarity, and pill-based contraception) may lead to the hypothesis that pregnancy denial could occur in a certain life context where pregnancy is impossible to be consciously apprehended by some women. Thus, to improve our interventions with them, medical professionals could focus on identifying and addressing those situational conditions.

To destigmatise women having experienced pregnancy denial, further research is necessary to confirm that newborns are not physically impacted.

Disclosures of interest
None declared. Completed disclosure of interest forms are available to view online as supporting information

Contribution to authorship
Conceptualisation: JA and ACR, with the contribution of CB (reagents/ materials/analysis tools). Acquisition of data: JA, ACR, ALSD, LV, VG, SSSG, ET, JLF, SV, GA, AD (psychiatrists); DD, DR, VM, AM, OP, ABM, MD, JBA, IN, OG (obstetricians). Data analysis: HD and AT. Data interpretation: HD, JE and ACR. Writing: HD. Supervision: ACR.

Details of ethics approval
The study received specific agreements from an independent ethics committee, the 'Comité de Protection des Personnes' (CPP) of Nancy, agreement no. 2011/56 on 7 May 2012, and has, therefore, been performed in accordance with the ethical standards laid down in an appropriate version of the Declaration of Helsinki of 1975, as revised in 2000. All participants provided informed written consent prior to their inclusion in the study.

Funding
This study was supported by grants from the Hospital Research Program of the French Ministry of Health (PHRC 2011). The funders had no role in the study design, data collection and analysis, decision to publish, or preparation of the manuscript.

Acknowledgements
At each centre, midwives, students and secretaries contributed to the smooth running of this study and we sincerely thank them. We also thank all the parents and infants who participated in this protocol.

Data availability
The datasets generated and analysed during this study are available from the corresponding author upon reasonable request.

Supporting Information

Additional supporting information may be found online in the Supporting Information section at the end of the article.

Appendix S1. Summary table of clinical studies on pregnancy denial. ■

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