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Long-term outcome of severe puerperal psychiatric illness: a 23 year follow-up study

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ABSTRACT

Background. Although there have been many follow-up studies of severe puerperal psychiatric illness, few have been very long-term.

Methods. Sixty-four subjects from 85 (75·3%) in an unselected sample of women admitted to a psychiatric hospital within 6 months of childbirth were successfully followed up a mean of 23 years (range 17–28) later. Most subjects were interviewed in detail, with further information obtained from general practice and hospital records. Data included subsequent illnesses and diagnoses, subsequent childbirth, longitudinal social function, current symptoms and social function.

Results. Seventy-five per cent of subjects had further psychiatric illnesses, most of them unrelated to childbirth, and 37% had at least three subsequent episodes. The risk of puerperal psychiatric illness was 29% in subsequent pregnancies. At outcome interview the majority of subjects were well, with satisfactory social adjustment. Diagnoses in subsequent psychiatric illnesses showed considerable consistency with index diagnoses, with some shift to bipolar disorder. Further illnesses were less likely to occur where the index illness occurred with first child, onset was within 1 month of delivery, and where the index diagnosis was unipolar depression.

Conclusions. There is a high risk of subsequent non-puerperal recurrences following severe puerperal psychotic illness, showing considerable diagnostic consistency with the index episode, but with good functional outcome. Puerperal illnesses showed strong continuities with non-puerperal illnesses in these women.

INTRODUCTION

Although there have been many follow-up studies of women with severe puerperal psychiatric illnesses, only two have been long-term (Protheroe, 1969; Rohde & Marneros, 1993). In addition to describing outcome, risk and predictors, follow-up studies, by considering nature of recurrences, can also answer questions as to nosology. Some authors have regarded severe puerperal illnesses as distinct nosological entities (Hamilton, 1962; Hays & Douglas, 1984; Hays, 1978), while others have considered them as episodes of affective or schizophrenic psychoses, occurring coincidentally in the puerperium or precipitated by it (Platz & Kendell, 1988).

Follow-up studies of unselected samples are difficult, since standard diagnostic systems do not separately identify puerperal episodes. Identified subjects have therefore tended to be those admitted to special mother-and-baby units. Also tracing is complicated because pregnancies occur at an age when subjects are geographically mobile, and because of surname change due to subsequent new marriages. We report a successful long-term 17–28 year follow-up taking advantage of a previously identified unselected sample of subjects admitted to the acute wards of a psychiatric hospital within 6 months of childbirth.

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METHOD

Subjects

Subjects were previously studied by Katona (1982). They were originally obtained by examining admission records to Fulbourn Hospital, Cambridge for 1970–1980 inclusive (excluding 1972 for which records were deficient). This hospital was then the sole NHS admission unit for a large catchment area in Cambridgeshire, and had no mother-and-baby unit. The discharge sheets were scrutinized to identify all women for whom the puerperium or an equivalent term was mentioned in diagnosis and onset within 6 months was established from case notes. Although ICD-8 was officially in use, these forms often used informal terms, that indicated the puerperium. This was supplemented by an examination of case records of other female admissions of appropriate characteristics (see Katona, 1982). Subjects for follow-up were 85 women, including all 84 original subjects and one additional subject hospitalized just outside 6 months from delivery, but whose symptoms had clearly begun earlier. The study was approved by the Cambridge Ethical Committee.

Tracing

Subjects were traced by NHS registration. The local Cambridgeshire Family Health Service Authority (FHSA) was first approached, and 46 subjects with local general practitioners were identified. The remaining 39 subjects were traced via the NHS Central Register through the Office for National Statistics (formerly the Office of Population Censuses and Surveys). It was possible to identify the current FHSA for subjects, and by enquiry to the FHSA, the currently registered GP. Deaths were also identified and copies of death draft entries obtained.

GPs were then contacted, requesting permission to contact the subject directly and the subject's address. Subjects were then approached by letter.

Interviews

Subjects were visited at home and interviewed by one of two researchers (S. A. R./V. J. D.), one a senior registrar in psychiatry and the other an experienced research assistant. Prior to the interview the original case notes were examined to obtain as much information as possible with regard to the index episode. Written consent for interview and use of case notes was obtained.

Information obtained

Illness episodes and diagnoses

In the original study (Katona, 1982) diagnoses from case notes had been made using DSM-III. As this has been replaced index episodes were rediagnosed and subsequent episodes diagnosed using the International Classification of Diseases, tenth revision (ICD-10 Research Criteria) (World Health Organization, 1993).

For follow-up a detailed semi-structured interview was devised, covering background information, index episode, previous and subsequent episodes, longitudinal aspects of social function, current symptoms and current social function. Efforts were made to identify all illness episodes. A screening interview identified key ICD-10 symptoms, and where a symptom was reported more detailed enquiry was made to establish a diagnosis. A new episode of mental disorder was coded when ICD-10 diagnostic criteria were fulfilled and there had been at least two intervening months symptom free. Information was collected for each episode, including index, concerning duration of symptoms, duration and type of treatment, age at onset, relation to childbirth, occurrence of a suicide attempt, in-patient or out-patient treatment and, if a puerperal episode, whether the subject had any thoughts of harming the baby, and also earlier family and life history. Information was verified and supplemented by inspecting GP records and hospital records of admissions from all hospitals concerned. For index episodes only, the presence was noted of the following atypical symptoms said to characterize severe puerperal illness (each coded as absent, mild/transient, or present to considerable degree): perplexity; misidentification of persons; disorientation; emotional lability; mood incongruent hallucinations or delusions; mood incongruent bizarre behaviour.

Current mental health was rated on a scale of 1 (well) – 7 (severely ill). Current social functioning was assessed using a modified version of the Social Adjustment Scale (SAS) (Weissman & Paykel, 1974). Background demographic information was also obtained.

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Longitudinal social functioning was rated on anchored scales developed for this study based on the Social Adjustment Scale (Weissman & Paykel, 1974). Details were also obtained of number of pregnancies and mental health of offspring. The latter will be reported separately.

Women who were traced to abroad were asked to return a postal questionnaire, comprising a modified form of the interview.

RESULTS

Tracing

Fig. 1 shows the results of tracing. Among the 85 subjects, 82 (96·5%) were traced (to current FHSA, special hospital, overseas or dead). Among the 82 traced subjects, four had died, 49 completed a full interview, in most cases at home, and one a detailed postal questionnaire providing comparable information. These 50 also allowed case note access. Fifteen women refused an interview or case note review, and in one further case the GP refused access. Twelve additional women refused interview but permitted access to GP records and subsequent hospital case notes for admissions. This information was also available for two women who had died. Thus, in 64 (75·3%) cases adequate information was available concerning psychiatric history after index episode either from interview, postal questionnaire or case records. In the remaining 21 (24·7%) cases no follow-up information was available other than, in two cases, that they had died. Thirty-four women had moved outside the Cambridge area and 21 subsequent admissions were to distant hospitals.

Among the four women who had died, a cause of death was identified in three of these. One died of pneumonia and two of self-inflicted poisoning, although only in one of these was suicide specified.

Sample characteristics

Table 1 comprises characteristics of subjects successfully followed up and those who were not. Only two variables showed significant differences. Those successfully followed up had longer duration of illness and a greater proportion had a previous puerperal illness. Information on those not followed up depended solely on index case notes and was sometimes incomplete or uncertain.

At the index episode most of the women later followed up were in their late 20s, with symptom onset within a month of delivery, about half in first pregnancies, about half with a previous psychiatric illnesses at some point, and one-quarter with a previous puerperal illness, including 16% whose previous illnesses were solely puerperal. The mean length of follow-up was 22·5 (S.D. 3·3) years, 18 subjects being followed up for 17–20 years, 28 for 21–24 years and 16 for 25–28 years.

A similar comparison was made between those interviewed and those with only case note follow-up information. Only two differences were significant, both at $P < 0{\cdot}05$. Interviewed

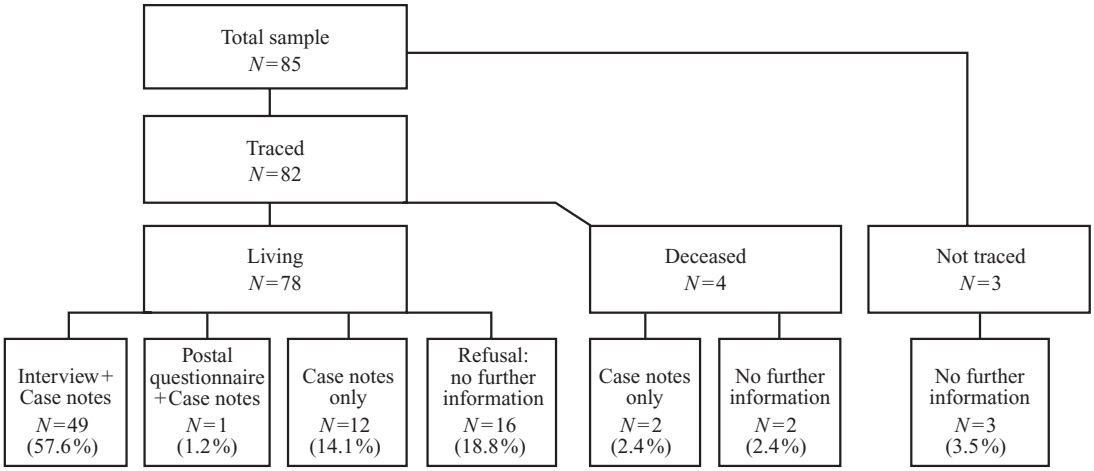


FIG. 1. Results of tracing and follow-up.

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Table 1. *Initial characteristics by sample*

	Follow-up ($N = 64$)		No follow-up information ($N = 21$)	
	N	%	*N*	%
Mean (S.D.) age at index episode (years)	28·0	(4·9)	27·1	(4·4)
Median (1st, 3rd quartile) duration of index episode (months)	5*	(2, 7)	2	(1, 4)
Mean (S.D.) duration of hospital admission (days)	44·5	(35·9)	41·6	(30·7)
Mean (S.D.) length of follow up	22·5	(3·3)	23·4	(4·2)
Marital status at index- married/cohabiting	61	95·3	20	95·2
Follow up by psychiatric services	47	73·4	14	66·7
Index onset within 1 month of delivery	46	71·9	15	71·4
Treatment at index episode with drugs	63	100·0	20	95·2
ECT	29	46·0	10	47·6
Psychotherapy	8	13·1	2	10·0
Index episode first pregnancy	28	43·8	11	52·4
Any previous psychiatric illness	33	51·6	3†	27·3
Any previous puerperal illness	17	26·6	0‡	0
Thoughts of harming child	14	25·0	2	11·8
Family psychiatric history	39	79·6	6	66·7

Data are presented as number and percentage unless otherwise indicated. Total *N*s differ among variables due to incomplete or uncertain data.

* $P = < 0{\cdot}01$ (Mann–Whitney test).

† Total *N* without follow-up information = 11.

‡ Total *N* without follow-up information = 15, $P = 0{\cdot}03$ two-tailed Fisher's Exact Test.

Table 2. *Diagnoses of index episodes*

	Follow-up ($N = 64$)		No follow-up information ($N = 21$)	
	N	%	*N*	%
Schizophrenia	3		1	
Schizoaffective	2		0	
Other psychoses	2		2	
Total schizophrenia and other psychoses	**7**	**10·9**	**3**	**14·3**
Mania	17		2	
Bipolar depressed (previous mania)	2		0	
Total bipolar disorder	**19**	**29·7**	**2**	**9·5**
Total unipolar depressive episode	**35**	**54·7**	**15**	**71·4**
Other diagnoses*	3		0	
Uncertain	0		1	
Total other/uncertain	**3**	**4·7**	**1**	**4·8**
Total	**64**		**21**	

* Two other depressive diagnoses, one somatoform autonomic dysfunction.

subjects were less likely to have had a previous psychiatric illness and more likely to receive ECT during the index episode.

Table 2 shows index diagnoses. Recoding diagnoses using ICD-10 did not lead to major change from Katona's study (Katona, 1982). Among the subjects followed up the most common diagnosis was of depressive episode or recurrent depressive disorder, with mania/bipolar disorder ranking second, and only seven schizophrenic or other psychotic diagnoses.

Subsequent illnesses

On follow-up 48 subjects (75%) had experienced subsequent episodes of psychiatric illness (38% one or two episodes, 37% three or more). The median number of further episodes was two. Twenty-nine per cent of the 34 subsequent live births after the index episode were associated with a further episode of illness. One woman, with thoughts of harming her children at index episode, had done so on follow-up.

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Table 3. *Outcome by index diagnosis*

		After index					
		Further episodes			Predominant diagnosis		
Index diagnosis		Any episode	Median no. of episodes	Median months to next episode	Schiz./psychotic	Mania/bipolar	Depressed
Schizophrenic/psychotic	*N* (7)	7	3	54	5	2	0
	%	100·0			71·4	28·6	0·0
Mania/bipolar	*N* (19)	17	2	38	0	17	0
	%	89·5			0·0	100·0	0·0
Depressed	*N* (35)	21	1	110	1	4	15
	%	60·0			5·0	20·0	75·0
Significance		< 0·02	NS	< 0·01			*

Excludes 'other' diagnosis ($N = 3$ at index, 1 after index) and those without follow-up information ($N = 21$).
Significance for medians tested by Kruskal–Wallis tests, otherwise by chi-square.
* Chi-square test not valid due to low cell frequencies.

Table 3 shows the occurrence of further illnesses, by index diagnosis. Subjects with an index diagnosis of unipolar depression were significantly less likely to have further episodes of illness than subjects with mania/bipolar disorder, or schizophrenia/other psychoses. They also had a significantly longer mean interval from index to first subsequent episode.

Table 3 also shows the subsequent diagnoses. For analysis these were grouped to generate a predominant diagnosis after index episode for each subject. Where diagnoses in episodes were mixed, schizophrenic diagnoses took precedence over affective. Depressive episodes with previous mania were classified as bipolar and took precedence over unipolar depression. There was considerable diagnostic stability on follow-up. Among the 47 subjects with further episodes, 37 had a subsequent predominant diagnosis in the same group as the index episode.

Diagnoses were also examined separately for each follow-up episode. Among the three subjects with index schizophrenia, all 11 subsequent episodes were diagnosed as schizophrenic. Among the two schizoaffective subject there were three subsequent schizophrenic episodes, two other psychotic and five manic episodes. Among the two other psychotic subjects there were seven subsequent schizophrenic episodes and three manic. Subsequent diagnoses in these groups were therefore predominantly schizophrenic but with a proportion of manic episodes.

Among the 17 subjects with index mania, there was much subsequent mania, with 55 subsequent manic episodes, six depressive episodes, one other diagnosis. The two index bipolar depressives had, in total, three subsequent depressive and two subsequent manic episodes.

Among the 35 index depressives, there were 44 subsequent depressive episodes and seven episodes with other diagnoses, but also 15 manic episodes in four subjects who therefore became bipolar.

Current outcome at time of interview

At time of interview the mean age was 50·2 years (S.D. 6·1) and 73% of subjects were married or in a stable cohabitation. At interview, 36 (72%) of the 50 subjects interviewed were rated as not mentally disordered at all, five as showing borderline mental illness, one as mild and only eight (16%) ill at moderate levels or above. However, 52% of 58 subjects on whom adequate information was available (including eight from case notes only) were currently receiving psychotropic medication.

Social adjustment

At the time of interview 71% of subjects were in employment (33% part-time, 38% full-time) and 29% were not working.

Current social adjustment was rated on five-point anchored item scales and seven-point global ratings of role areas, from the Social Adjustment Scale (Weissman & Paykel, 1974). Social adjustment was generally good on the five-point item scales, with the majority of scales showing 70–80% of subjects at point 1 (very good function) and 90% or more at either point

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Table 4. *Predictors of further episodes*

Predictors	Any further episodes after index: No *N*	No %	Yes *N*	Yes %	χ^2
Family history of mental illness					
No	3	30·0	7	70·0	0·00
Yes	11	28·2	28	71·8	
Quality of childhood					
Good/fair	8	27·6	21	72·4	0·00
Poor/hostile	10	31·3	22	66·9	
Relationship of subject with mother during childhood					
Good/fair	9	26·5	25	73·5	0·02
Poor/hostile	3	20·0	12	80·0	
Any episode before index					
No	9	29·0	22	71·0	0·19
Yes	7	21·2	26	78·8	
Any pregnancies before index					
No	11	39·3	17	60·7	4·15*
Yes	5	13·9	31	86·1	
Age at index episode (years)					
19–25	6	27·3	16	72·7	2·12
26–29	7	33·3	14	66·7	
⩾ 30	3	14·3	18	85·7	
Index onset					
> 1 month of delivery or during pregnancy	0	0·0	18	100·0	6·60**
within 1 month of delivery	16	34·8	30	65·2	
Duration of index episode					
0–4 months	6	24·0	19	76·0	0·10
> 4 months	10	31·3	22	68·8	
Atypical symptom present, at least transiently, in index episode					
No	11	26·2	31	73·8	0·00
Yes	5	23·8	16	76·2	

Percentages are by row. Total *N*s differ among variables due to incomplete or uncertain data.
* $P < 0·05$; ** $P < 0·01$.

1 or 2 (normal function). On 1–7 global ratings for work, social and leisure activities, marital relationships, parental role, 75–85% rated at point 3 (mild maladjustment) or better. On an overall global rating of current adjustment, 15% of subjects were rated as excellent, 45% as good, 19% as mildly maladjusted and only 21% showing moderate or greater maladjustment.

For longitudinal social function detailed enquiry was made concerning: work history (number of full-time and part-time jobs, longest time in one job, reasons for changing jobs, pattern of work in relation to children, reasons for not working); marital relationships (length of current marriage, length of longest cohabitation, number of previous separations/divorces, violence in marital relationships); parental history (ability to provide and care for children emotionally and physically). On the basis of these items global ratings were made on four or five-point scales. On these scales 67% of women rated as having good or adequate employment histories, 18% deficiencies and only 14% very poor employment. Sixty-eight per cent were rated as having good or adequate longitudinal marital adjustment, 23% as showing deficiencies and 9% very poor adjustment. Thirty-one per cent of those ever married had a marriage end in divorce or separation, and 14% had two such marriages. Seventy per cent had shown good or adequate parental function, 22% deficiencies and 8% very poor parental function.

Predictor analyses

A number of predictor variables were cross tabulated against the occurrence of any further psychiatric episodes, the findings are shown in Table 4. Further episodes had already been found to be significantly less likely to occur

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where the index diagnosis was unipolar depression (Table 3). In addition, further episodes were less likely to occur where the index pregnancy was the first pregnancy, and where index onset was within 1 month after delivery. No other variables predicted further illnesses. There were too few puerperal recurrences for predictors of these, including index diagnosis, to be examined.

The three significant predictors from index episode were also cross-tabulated with each other. Early puerperal onset was more likely to occur following the first pregnancy, but diagnosis was not related to early onset or first pregnancy. Atypical symptoms, which did not predict outcome, were also not associated with any of the three significant predictors.

DISCUSSION

Previous studies

Follow-up studies of puerperal illnesses can answer clinically relevant questions concerning risks of puerperal and non-puerperal recurrence and prognostic factors, and can also address the nature of post-partum disorders through the clinical pictures of non-puerperal recurrences. Very long-term follow-ups, such as the present study, are required to produce accurate findings with regard to non-puerperal recurrences and their clinical features, and also functional outcomes.

There have been a number of previous studies. Those with follow-up periods of up to 6 years include Martin (1958), Kadrmas *et al.* (1979), Da Silva & Johnstone (1981), while those between 6 and 15 years include Davidson & Robertson (1985), Platz & Kendell (1988), Bell *et al.* (1994), Videbech & Gouliaeve (1995). These shorter follow-up studies are sufficient to cover the main childbearing years and have indicated recurrence rates around 1:4 or 1:5 in subsequent pregnancies (Martin, 1958; Davidson & Robertson, 1985). Some of these studies also suggest high recurrence rates outside the puerperium, for instance 43% over 8–15 years in unipolar depressive puerperal illnesses (Davidson & Robertson, 1985). Recurrence rates appear lower after puerperal illnesses than for diagnosis-matched non-puerperal illnesses (Platz & Kendell, 1988; Dean *et al.* 1989).

Two studies have extended beyond 15 years, both of disorder of onset within 6 weeks of delivery. Protheroe (1969) interviewed 85 women a mean of 15 years (range 4–35) after admission. Forty-seven per cent of subjects had at least one further illness, mainly non-puerperal. At the time of follow-up, 86% of subjects were symptom free. Diagnoses in subsequent illnesses were nearly always the same as in the index illness. Few other studies have addressed diagnostic stability. Rohde & Marneros (1993) studied 61 women after a mean of 26 years (range 12–41). A total of 64% had at least one recurrence, most often several. Seventy-two per cent were symptom-free at follow-up. Findings of these studies were very similar to our own.

Few studies have examined social function. Videvech & Gouliaeve (1995) found that 40% of women had not preserved full working capacity, due to mental illness, even though most were not schizophrenic. However, Rohde & Marneros (1993) reported good outcome on the WHO 6-point Disability Assessment Schedule, with 72% showing good adjustment and a further 18% satisfactory adjustment. On the Global Adjustment Scale 72% scored 91–100.

Uncertainty exists regarding prognostic factors. Dean *et al.* (1989) found that subjects with no previous non-puerperal illness had the best outcome. Protheroe (1969) found better outcome following manic depressive than schizophrenic disorders.

Findings and implications

This follow-up study of women with hospitalized puerperal illnesses has found high recurrence: 75% of subjects had a further psychiatric illness, mostly non-puerperal, and half of these had at least three episodes. Diagnosis showed considerable consistency with the index puerperal episode. Psychiatric and functional state at the end of the study and lifespan adjustment were relatively good, but many subjects were still on psychotropic drugs. These disorders were episodic rather than chronic or deteriorating.

A recent Danish study (Appleby *et al.* 1998) reported a high suicide SMR of 1719 in puerperal disorders. The present sample of 85 is small but the occurrence of two suicides over an average of 23 years is comparable with that expectation (approximately three).

The sample was an unselected one from admissions to a catchment area psychiatric

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hospital. Some eligible subjects might have been missed if patient lists did not refer in some way to the puerperium. An estimate of expected numbers can be derived from the total catchment area population served (approximately 550000). An annual birth rate of 1·4%, and incidence of severe psychiatric disorder of 1:1000 or 1:750 births, would produce expected numbers between 77 and 103, compared with 85 subjects studied.

Tracing was almost complete. There were some refusals, but given the long interval, with index illnesses not always known to subsequent partners, these were comparatively few. There may have been some selection for worse outcome on index characteristics of those interviewed, but this was not major. In about half our subjects the first illness was the index episode and in 16% more the only previous illnesses had been puerperal.

We were unable to follow-up comparison non-puerperal psychiatric cases so we cannot directly conclude as to whether outcome is better for puerperal disorders, as other shorter follow-up studies suggest. We primarily aimed to provide good long-term follow-up information for puerperal disorders. Our findings for the predominant subjects with affective disorder do appear better than in the major longer term follow-up studies of Lee & Murray (1988) and Kiloh *et al*. (1988). Findings on social adjustment at interview were slightly worse than those found by Weissman & Paykel (1974) in US normals. Neither comparison may be valid. There are no comparable data for longitudinal social adjustment.

Although some severe post-partum illnesses are typical mania, depression or schizophrenia, some, particularly with early onset, show atypical features including labile mood depression and elevation, schizophreniform symptoms, atypical delusions, and confusion (Protheroe, 1969; Hays, 1978; Kadrmas *et al*. 1979; Brockington *et al*. 1981; Dean & Kendell, 1981; Wisner *et al*. 1994; Oosthuizen *et al*. 1995). In the English language literature briefly reviewed in this paper, the debate has particularly concerned these and whether they represent bipolar disorder, or a specific disorder (Hays, 1978; Hays & Douglas, 1984). There is also good evidence of raised rates of typical mania in the puerperium.

Aetiology of the severe puerperal illnesses is not clear. They are not much related to life stress (Brockington *et al*. 1989; Dowlatshahi & Paykel, 1990; Marks *et al*. 1991). Family, history studies are conflicting (Kadrmas *et al*. 1979; Platz & Kendell, 1988; Schopf *et al*. 1985; Dean *et al*. 1989). There is some evidence of dopamine receptor abnormalities preceding onset (Wieck *et al*. 1991) and of raised cortisol post-dexamethasone during illness, a state marker consistent with affective disorder (Paykel *et al*. 1991).

Follow-up studies may, therefore, throw light on the diagnostic question. Diagnoses in the present study were predominantly affective, with a high rate of mania, consistent with other studies. Atypical symptoms were spread across diagnoses and did not predict outcome. Our findings of better prognosis of episodes with early puerperal onset and episodes associated with first pregnancy suggest that particular special characteristics are associated with these. On the other hand the subsequent diagnostic consistency points to strong continuities with non-puerperal psychiatric disorder. The good functional outcome, in spite of high recurrence rates, and continued medication, is consistent with affective disorder. Direct inferences cannot be made, but the suggestive pointers are towards a disorder, predominantly affective in nature, precipitated by the puerperal state, perhaps by biological factors and with a moderate tendency to recurrence in the same form.

The study also has implications for the clinician. Shorter follow-up studies have established the risk of illness in subsequent childbirth. Our findings on much longer follow-up indicate that puerperal illness must be taken seriously as indicating comparatively high risk of further non-puerperal episodes of recurrent psychiatric disorder, although with good remission and functional outcome.

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