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Follow-up and family study of postpartum psychoses**Part III: characteristics of psychoses occurring exclusively in relation to childbirth**

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Abstract As part of a follow-up and family study of postpartum psychoses, this episode of illness being the first leading to psychiatric hospitalisation, patients with puerperal episodes (PE) and nonpuerperal episodes (NPE) of illness in the long-term course ($n = 79$) were compared to patients with PE only ($n = 40$). Few differences were found. Relatives of patients with PE only had a lower morbidity risk for functional psychoses than relatives of patients with PE and NPE. A favourable course of illness in the presence of a low genetic predisposition may be expected, according to the diathesis-stress model of functional psychoses.

Key words Parity · Genetics · Diathesis-stress model

Introduction

Until the 19th century many authors had the opinion that severe psychiatric postpartum disorders were caused by complications of delivery, puerperium or lactation. For example, milk was believed to penetrate the brain, distend the brain fibres and in this way to provoke mental disturbances. However, already Esquirol (1838) raised doubts concerning this aetiology.

Kraepelin (1920) and Bleuler (1916) classified most of the postpartum psychoses among the endogenous (functional) psychoses. Organic psychoses were also frequently diagnosed in the 20th century (Thomas and Gordon 1959), but this was often done based on simple fever in the puerperium and without any other definite evidence of brain pathology. In the past decades organic postpartum psychoses have definitely become rare (Protheroe 1969; Rehman et al. 1990).

Postpartum psychoses are considered functional psychoses starting in a particular period. Many patients suffering from such disorders also have psychotic episodes un-

related to childbirth, and the family history is often positive for functional psychoses (Schöpf 1994). Nevertheless, some investigators continue to believe that at least some postpartum psychoses represent a distinct group aetiologically linked to events of the puerperium, with signs of confusion considered characteristic (Hamilton 1962, 1982).

According to the knowledge of the authors, few studies on psychoses occurring exclusively in the postpartum period have been performed. McNeil (1988) compared women with puerperal episodes (PE) only, women with PE and nonpuerperal episodes (NPE) and childbearing women with NPE only. He found some differences, e.g. a higher social class and a shorter hospitalisation time in the first group, however, no statistical comparison of the very small samples in the diagnostic subgroups was performed. McNeil (1988) also reported differences in the liability of PE for successive deliveries. The present follow-up and family study compares characteristics of patients with PE only vs patients with PE and NPE.

Method

As described in detail elsewhere (Schöpf and Rust 1994), 119 patients with postpartum psychosis and 542 first-degree relatives were investigated. The index episode began within 3 months following delivery and was the first decompensation leading to psychiatric hospitalisation. The patients were admitted to the Psychiatric University Hospital of Lausanne between 1949 and 1980 or to the Psychiatric University Hospital of Zurich between 1956 and 1964, and followed up after a mean of 21 years (range 2–35 years). Patients were classified using the DSM-III-R. Psychotic episodes of illness unrelated to childbirth, including depressive episodes with incapacitating features, melancholic type according to the DSM-III-R, were considered NPE.

Results

A comparison of the two groups is made in Table 1. No statistically significant differences were found for characteristics of the index episode. Notably, the proportion of confusio-oid syndrome did not differ in the two groups. There were differences in the long-term course, but these were related to the classification criteria of NPE.

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Table 1 Characteristics of patients with PE only compared to patients with PE and NPE (nominal values in %, quantitative data $x \pm s$). For the exact definitions of items see Schöpf and Rust (1994). (*D* depression; *M* mania; *MSCH* schizoaffective mania;

DSCH schizoaffective depression; *SCHF* schizophreniform psychosis; *BRP* brief reaction psychosis; *SCH* schizophrenia; *SCHA* schizoaffective psychoses; *A* affective psychosis)

	PE only (<i>n</i> = 40)	PE and NPE (<i>n</i> = 79)	<i>p</i> -value	Correlation
<i>Index episode, personal history</i>				
DSM-III-R diagnoses:				
D/M, MSCH/DSCH/SCHF, BRP/SCH	47.5/12.5/0/37.5/2.5	57/16.5/9/16.5/1	† (1)	0.28 (5)
Cycloid psychosis	27.5	32	n.s. (1)	0.04 (5)
Confuso-oneiroid syndrome	30	32	n.s. (1)	0.02 (5)
Paranoid syndrome	2.5	16	n.s. (4)	0.19 (5)
Acute onset	52.5	43	n.s. (1)	0.09 (5)
Early onset	75	76	n.s. (1)	0.01 (5)
Age at delivery	27.2 \pm 4.9	26.8 \pm 4.8	n.s. (3)	0.04 (6)
Primiparae	65	58	n.s. (1)	0.04 (5)
Psychopathology during index gravidity	20	20	n.s. (1)	0.00 (5)
Previous psychotic episodes	0	19	0.004 (1)	0.27 (5)
Age of first episode of illness	27.1 \pm 4.9	25.8 \pm 4.9	n.s. (3)	0.13 (6)
<i>Follow-up</i>				
DSM-III-R diagnoses:				
A/SCHA/SCHF, BRP/SCH	65/2.5/30/2.5	53/25/9/13	† (1)	0.40 (5)
Global psychopathological outcome	0.3 \pm 0.5	1.7 \pm 0.7	0.001 ^a (2)	††
Duration of follow-up (duration of survival in patients who had died)	21.3 \pm 9.4	21.1 \pm 8.1	n.s. (3)	0.00 (6)
<i>Family history</i>				
Family history of functional psychoses	12.5	46	0.001 (1)	0.33 (5)
Family history of functional psychoses, index episode	7.5	35	0.002 (1)	0.30 (5)
Global morbidity risk	5.3	13.2	0.02 (1)	0.11 (5)

^a Approximate measure (ordinal scale niveau)

† = The global χ^2 test may not be applied reliably due to expected frequency <5 in >20% of cells. †† = Due to variance hetero-

geneity the effect size could not be determined. *Statistical tests:* (1) χ^2 ; (2) *u*-test; (3) *t*-test; (4) Fischer-Yates test; (5) phi coefficient; (6) point-biserial correlation coefficient

The family history for functional psychoses was significantly more often positive in patients with PE and NPE (Table 1), with this difference already being present at the index episode. Moreover, the global morbidity risk for functional psychoses differed significantly.

The secondary cases were evaluated for two characteristics which may tentatively be considered indicators of the degree of hereditary predisposition for functional psychoses. The age of onset was 35.3 ± 11.4 years in the secondary cases of patients with PE and NPE, and 28.7 ± 19.0 years in the secondary cases of patients with PE only (n.s.). The proportion of secondary cases with the other characteristic, i.e. schizophrenic symptoms, including mood-incongruent psychotic features, was 38% and 71%, respectively, in the two groups (n.s.).

The classification according to the presence of NPE was also performed in the 42 patients who had further children and could be evaluated concerning the occurrence of puerperal relapses (Schöpf and Rust 1994). In the patients with PE only the frequency of a puerperal relapse after the next delivery was 4 of 18 (22%), and in the patients with PE and NPE, 12 of 24 (50%). Because the risk of relapse might be related to parity, the two groups were

controlled for this characteristic. At the index episode 12 of 18 patients (67%) and 18 of 24 patients (75%), respectively, were primiparae. The comparison of the 18 patients of the first group with matched patients of the second group resulted in a proportion of puerperal relapses of 22% and 44%, respectively (n.s.).

Discussion

Among negative results found in this study, the proportion of a confuso-oneiroid syndrome did not vary in the two groups. This does not support the assumption of Hamilton (1962, 1982) that signs of delirium are a special characteristic of psychoses occurring exclusively in the postpartum period.

In this investigation the first-degree relatives of patients with PE only had a lower global morbidity risk for functional psychoses than relatives of patients who had NPE. Because the relatives were not systematically interviewed, this finding might represent an artefact. In both groups one principal source was interviewed on the family history, but in patients with PE only there were fewer sources of information and therefore fewer possibilities to ascertain

the completeness of the data. However, a significant difference in the family history was already present at the index episode when the sources of information were the same.

The difference in global morbidity risk for functional psychoses in relatives of subgroups of patients classified according to the presence of NPE has already been reported in a previous investigation in the Lausanne sample (Schöpf et al. 1985). In the patients hospitalised in Zurich there was a minimal and nonsignificant difference in the same direction (12.8% vs 11.9%; Schöpf 1994). Considering the total sample it appears justifiable to assume that there is a difference. In a study by Ifibamuyi and Akindele (1985) a positive family history was found in 8 of 36 patients with PE only compared to 9 of 14 in patients with PE and NPE ($\chi^2 = 7.948, p < 0.006$). However, Dean et al. (1989) compared the morbidity risks in relatives of patients with NPE and relatives of patients without NPE and found no difference.

The relation between family history and long-term course is compatible with the diathesis-stress model of functional psychoses. The birth of a child is possibly the strongest provoking event for psychoses. Kendell et al. (1987) found that the psychiatric hospitalisation rate was 20-fold higher in the 4 weeks after delivery than outside the postpartum period. It appears plausible that women with a low genetical predisposition may develop a psychosis in the postpartum period, and not fall ill following psychosis-provoking events of lesser severity or in their absence.

The affected relatives of patients with PE only did not have a higher age at onset or higher frequency of schizophrenic symptoms, including mood-incongruent psychotic features, than relatives of patients with PE and NPE. Both variables were used as possible indicators of the genetic predisposition for functional psychoses. There is a relation between early age at onset and positive family history (Gershon et al. 1976; Baron et al. 1981). The appearance of schizophrenic symptoms in a patient is determined by the genetic predisposition to schizophrenia, but probably also by the severity of disease milder forms sometimes manifesting only with affective symptoms. For example, schizoaffective psychosis has been called a virulent form of affective psychosis (Gershon et al. 1982). The lack of difference in the two characteristics might indicate that in the group of patients who had only PE, patients with an affected relative had the usual genetic predisposition, whereas patients without an affected relative had no genetic predisposition. This would mean that the latter are aetiologically unrelated to the known groups of functional psychoses. Traditionally one assumes that there is a genetic predisposition also in patients without positive family history. An absence of manifest illness in relatives in a proportion of families is expected in polygenic inheritance and incomplete penetrance, the kind of genetic transmission likely to be present in functional psychoses. If the hereditary predisposition was homogeneously distributed in the group of patients with PE only, an increased age at onset and/or a decreased proportion of cases with schizophrenic symptoms should have occurred.

In an investigation by McNeil (1988), the frequency of PE in the patients who had NPE remained constant at ap-

proximately 25% for successive deliveries, whereas patients with PE only showed a frequency of PE decreasing from 89% (first child) to 33% (second child) to 0% (third child). McNeil postulated a special relationship to primiparity of the postpartum-only group. The results of the present study appear consistent with this finding, because patients with PE only showed a (nonsignificant) lower proportion of puerperal relapse following the next delivery than patients with PE and NPE. However, one might question whether the conclusion made by McNeil can definitely be drawn from his findings. He showed the decreasing frequency of PE in patients with PE and no NPE. It cannot be excluded also that patients with NPE had a decreasing vulnerability to PE, but that the low frequency of an episode of illness after the first child resulted from a confounding influence. Some patients had their first child before the onset of psychosis of nonpuerperal type, when possibly the factors predisposing to psychotic breakdown were not yet present. The low frequency of puerperal episodes of approximately 25% is compatible with this possibility.

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