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On the Nosology of Severe Psychiatric Post-Partum Disorders

Results of a Catamnestic Investigation

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Summary. A group of 57 women, who had been hospitalised for puerperal psychiatric disorders from 1958 to 1977, were reexamined in 1982. The aim of the study was to determine the proportion of patients who had suffered from nonpuerperal psychotic relapses or other subsequent psychopathology, to define the sample diagnostically, taking into account progress in classification, to characterize the so far relatively neglected later course of illness, and to establish criteria related to relapse and global clinical outcome.

Of these patients 65% had at least one nonpuerperal relapse, only 25% remained free of later psychopathology, but the global outcome was favorable or relatively favorable in many cases. Of the patients who had had nonpuerperal relapses 43% were classified as suffering from affective psychosis, as many as 38% from schizoaffective psychosis, and only 19% from schizophrenia. Schizoaffective psychosis seems to be particularly liable to be provoked by childbirth. No major evidence was found that endogenous psychoses with puerperal onset and nonpuerperal relapses have a course of illness different from that of the corresponding diagnostic category in general. Cases with exclusively puerperal decompensations seem to be nosologically independent from the traditionally recognized endogenous psychoses. Characteristics strongly related to nonpuerperal relapses were a family history of psychosis and the occurrence of psychotic episodes before the index episode. Puerperal relapses occurred at a much higher rate in patients who also had nonpuerperal relapses than in patients without.

Key words: Nosology – Puerperal psychoses – Follow up – Schizoaffective psychosis

Zusammenfassung. Eine Gruppe von 57 Frauen, die in der Zeit von 1958 bis 1977 wegen psychischer Störungen im Puerperium hospitalisiert worden waren, wurde im Jahr 1982 nachuntersucht. Ziel der Studie war es, den Anteil von Patientinnen mit nichtpuerperalen psychotischen Rückfällen oder anderer im späteren Leben auftretender Psychopathologie zu ermitteln, die Patientinnen unter Berücksichtigung neuerer Fortschritte der psychiatrischen Klassifikation diagnostisch zu charakterisieren, den bisher wenig untersuchten weiteren Krankheitsverlauf zu beschreiben und prognostische Kriterien für Rückfälle und den weiteren globalen klinischen Verlauf zu erstellen.

65% der Patientinnen hatten nichtpuerperale Rückfälle, und nur 25% blieben frei von jeglicher Psychopathologie, jedoch der globale klinische Verlauf war in vielen Fällen günstig oder relativ günstig. Bei 43% der Patientinnen mit nichtpuerperalen Rückfällen wurde die Diagnose einer Affektpsychose, bei 38% die einer schizoaffektiven Psychose und bei nur 19% die einer Schizophrenie gestellt. Schizoaffektive Psychosen scheinen besonders häufig durch die Geburt eines Kindes ausgelöst zu werden. Es wurden keine wesentlichen Hinweise dafür gefunden, daß endogene Psychosen mit puerperalem Beginn und nichtpuerperalen Rückfällen einen anderen langfristigen Krankheitsverlauf haben als die entsprechende diagnostische Kategorie im allgemeinen. Fälle von ausschließlich puerperalen Dekompensationen scheinen von den traditionell anerkannten endogenen Psychosen nosologisch unabhängig zu sein. Mit nichtpuerperalen Rückfällen korrelierte Charakteristika waren eine Familienanamnese von Psychosen und das Auftreten von psychotischen Störungen vor der Indexerkrankung. Puerperale Rückfälle häuften sich unter den Patientinnen, die auch nichtpuerperale Rückfälle hatten.

Schlüsselwörter: Nosologie – Puerperalpsychose – Katamnese – Schizoaffektive Psychose

Introduction

By post-partum period or puerperium one means an unspecifically defined span of weeks or months after confinement. This period makes patients vulnerable to psychopathology of various types and degrees of intensity. Half or more of the mothers have a short episode of tearfulness a few days after delivery, the so-called post-partum blues (Robin 1962; Yalom et al. 1968; Pitt 1973). Frequently, longer lasting depressive states develop, in general of moderate intensity, with an incidence of depression of 10% or more (Pitt 1968; Kumar and Robson 1978; Paykel et al. 1980; Cox et al. 1982). Severe post-partum disorders, which are often summarized under the term "post-partum psychosis" or "puerperal psychosis", have been found by several authors to occur no more than twice per 1000 confinements (Hemphill 1952; Osterman 1963; Paffenbarger 1964; Grundy and Roberts 1975; Kendell et al. 1981). This rate, although low, means a sharp rise in the incidence of functional psychoses in comparison with periods unrelated to childbirth (Pugh et al. 1963; Kendell et al. 1976, 1981; Nott 1982).

Post-partum psychosis is no longer considered as a disease entity with its own psychopathology and course. However, "atypical" characteristics in puerperal psychoses have often been described (Marcé 1858; Fürstner 1875; Ripping 1877;

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Table 1. Follow-up studies of severe post-partum disorders

Author	Size of original sample	Duration of follow-up	Diagnostic classification of post-partum disorder: affective / "schizoaffective" / schizophrenic / other	Percentage of patients with – nonpuerperal relapse or chron. psychot. evolution[a] – poor outcome[b]
Schneider (1957)	28	2.0–15.0 years	– – – –	22[a]
Foundeur et al. (1957)	100	0.7–10.0 years	25 – 50 25	28[b]
Martin (1958)	75	0.3–10.5 years	37 39 20 4	9[a]
Saeger (1960)	42	0.5– 5.0 years	55 – 30 15	27[a]
Jansson (1964)	195	6.2–11.3 years	– – – –	29[a]
Arentsen (1968)	168	6.0–30.0 years	24 – 11 65	44[a]
Protheroe (1969)	134	1.0–35.0 years	68 – 27 5	39[a]
Wilson et al. (1972)	44	1.0–15.0 years	30 – 57 23	26[b]
Da Silva and Johnstone (1981)	47	1.0– 6.0 years	47 – 40 13	49[b]

Savage 1875; Kluge 1942; Hamilton 1962; Brockington et al. 1981; Katona 1982). This is evidence that they do represent a specific selection of psychoses and not merely a cross section of endogenous psychoses of women of child bearing age.

Even before the introduction of modern psychotropic drugs, it had been observed that only approximately 15% of severe post-partum disorders developed into chronic incapacitating disorders (Schneider 1957). The risk of further puerperal relapses has been found to be about one in seven for each subsequent pregnancy by a number of authors (Martin 1958; Arentsen 1968; Protheroe 1969) but a higher proportion of one in three has also been found (Paffenbarger 1964).

Several studies concentrating on the investigation of nonpuerperal psychopathology, in particular of nonpuerperal psychotic relapses, in later life have been performed (see Table 1), but only the investigations of Arentsen (1968) and Protheroe (1969) covered a long enough observation period to determine the proportion of cases with psychotic episodes, unrelated to childbirth in later life; even in Protheroe's study, the observation time was short in a number of cases. A limitation in all follow-up studies, except for that of Da Silva and Johnstone (1981), is the lack of defined diagnostic criteria. The problem is illustrated by the great variation in the distribution of diagnoses for the puerperal episode between the different investigations, which have all been performed on nonselected samples. Furthermore, important aspects of later clinical evolution, like the course of illness in patients with relapses, has so far not been investigated thoroughly.

The aim of the present investigation was first to investigate the further evolution of patients with severe post-partum disorders in a sample with a long catamnestic observation period in respect to further psychotic episodes, nonpsychotic psychopathology and global clinical outcome; secondly, to characterize the long-term evolution of these patients benefiting from progress in classification, i.e., by differentiating unipolar endogenous depression from bipolar manic depressive psychosis and by admitting that there is such an entity as schizoaffective psychosis which—at least according to our present knowledge—can neither be assigned to affective psychosis nor to schizophrenia. The evaluation was made using operationalized diagnostic criteria. Thirdly, we tried to find characteristics related to puerperal and nonpuerperal relapses or an unfavorable evolution in general.

Method

All women admitted to the Psychiatric University Clinic of Prilly-Lausanne for a post-partum disorder in the years 1958–1977 were eligible. The clinic has functioned as a sector hospital since 1961, and this sample can thus be considered representative for post-partum disorders requiring psychiatric hospitalisation. The following inclusion criteria were established: (1) the index hospitalisation had to be the first major psychic decompensation in adult age, i.e., the first episode of illness requiring psychiatric hospitalisation, (2) the beginning of the illness had to occur within 3 months after confinement, and (3) the patient must not suffer from marked mental retardation corresponding to an IQ of 70 or less, from epilepsy or other chronic neurological brain diseases. The cases were identified by a preexisting list, which had already been used for a clinical study of post-partum disorders (Jonquière 1981). In this way, 61 patients were identified.

In a first step, background information was gathered and the clinical picture of the post-partum episode was investigated using the case records. The second step, i.e., the follow-up investigation, was performed between January and October, 1982; this means that the time since the post-partum episode was 5–24 years. We searched for information about further psychopathology, the treatment received, the social functioning, and any new mental diseases in first degree relatives. Most patients were contacted by telephone and were asked for a personal interview. When we learned that a woman was still undergoing psychiatric treatment, we asked the psychiatrist whether he considered the interview as contra-indicated; in such cases, he gave us the necessary information. Moreover, we consulted all case records of other institutions concerning the patient and her first degree relatives. Sometimes, we obtained additional information from general practitioners, the husband or from other relatives. Among the 61 patients, we were unable to locate 3 foreigners who had returned to their countries, and 1 patient refused to participate in the study. The final sample consists, therefore, of 57 patients, which is 93% of the original group. In the study 5 patients, who committed suicide several years after the index episode, were included since detailed information on their further evolution was available. We had a personal interview of 1 to 2 h with 32 patients, usually in their homes. In the remaining 20 cases, we had tele-

phone interviews with 16 women, who were too far away for direct contact; 4 patients were not directly contacted.

For diagnostic classification, two main principles were respected: (1) the distinction between the diagnosis of episodes of illness and long-term diagnosis, and (2) the establishment of long-term diagnosis excluding the puerperal episodes. The former principle is particularly important for diagnosis of schizoaffective psychosis, which, for our purposes, is considered exclusively as a long-term diagnosis. In its evolution, purely affective, purely schizophrenic, and mixed affective-schizophrenic episodes can occur (Angst et al. 1979a). The latter was respected since the possibility was considered that post-partum episodes might have unique clinical characteristics which never occur outside the puerperium. The expression of a "nonpuerperal relapse" was limited to psychotic episodes.

For classification of puerperal episodes, nonpuerperal relapses and psychotic episodes before the index one not leading to hospitalisation, five categories were established: depression, mania, depression or mania with schizophrenic symptoms, and nonaffective psychotic episode. Definitions were derived from the Research Diagnostic Criteria (RDC) (Spitzer et al. 1978). Depressions, defined as major depressive disorders, were only considered as present if they fulfilled the level "definite", and pure depressions were evaluated for endogenous features (see below). In our terminology, we did not use the expression "schizoaffective" for episodes of illness, in order to avoid confusion with long-term diagnosis. In the diagnosis of a nonaffective psychotic episode, we included all episodes of illness with an RDC diagnosis of a schizophrenic episode or an episode of unspecified functional psychosis. Furthermore, we made the following modifications to correspond to our understanding of the ICD diagnoses: (1) the definition of depressive or manic episodes with schizophrenic symptoms, differentiated from episodes with pure depression or mania, was wider than the RDC diagnoses for schizoaffective episodes, as it included all episodes in wich clearcut mood-incongruent delusions or hallucinations occurred; (2) major depressions occurring with schizophrenic symptoms were only included in the diagnosis if they occurred in the acute phase of the illness; (3) for depressions, the terms "endogenous" and "psychotic" were used synonymously. We considered as endogenous depressions all episodes that fulfilled the RDC criteria of an endogenous subtype as well as episodes with depressive delusions or hallucinations or marked and persistent motor inhibition/agitation; in addition, they had to be accompanied by a marked diminution of working capacity. As far as depressive post-partum episodes are concerned, we finally abandoned the attempt to differentiate endogenous from non-endogenous ones, since there were several patients with a doubtful endogenous symptom profile, whom it would have been arbitrary to classify in either of the two groups. The classification of nonpuerperal depressions was much easier, and we therefore applied the distinction. If a patient had had one relapse with an endogenous depression, we considered all other depressive nonpuerperal episodes of illness also as endogenous, unless there was major evidence of a purely reactive disturbance, such as a transitory urge to suicide immediately after separation from a husband, etc. The diagnostic classification of the post-partum episode was made by two authors (J. Sch.; C.B.) independently. In case of disagreement, all available documents were reviewed, to arrive at a definite diagnosis.

Nonpuerperal episodes of hypomania according to the RDC were not considered as relapses; however, they contributed to establish the long-term diagnosis of manic depressive psychosis and if they lasted 2 weeks or longer the determination of the cycle length (see below). For classification of non-psychotic psychopathology, the RDC were also used.

According to the unmodified RDC, all psychiatric disturbances with onset of illness within one week after childbirth or with disorientation are considered as of organic origin. We classified only those cases as suffering from an organic psychosis, in whom somatic evidence for an organic etiology was found.

In all patients with nonpuerperal relapses, one of the following long-term diagnoses was made: (1) unipolar endogenous depression, (2) bipolar manic depressive psychosis, (3) schizoaffective psychosis, and (4) schizophrenia. All cases corresponding to bipolars I or II, according to the RDC, were included in the second group. Schizoaffective psychosis was differentiated from affective psychosis in such a manner that patients who had once had a nonpuerperal episode with schizophrenic symptoms, with or without an affective syndrome, were no longer considered as suffering from affective psychosis. The distinction from schizophrenia was made in the following way: patients were classified as schizoaffective if, for the majority of the episodes, a diagnosis with an affective syndrome (pure or with schizophrenic symptoms) was made. In addition, the long-term course of the illness had to show a certain periodicity. In the long-term evolution of a case however, if schizophrenic symptoms with no affective component dominated, e.g., if there were only occasional affective swings superimposed on chronic nonaffective psychotic symptoms, then the patient was considered as suffering from schizophrenia in the long-term diagnosis.

Results

Background Data and Clinical Characteristics of Post-Partum Illness

The 61 patients represented two-thirds of all admissions for puerperal disorders to the psychiatric clinic during the 20-year period. The proportion of post-partum disorders among all female admissions was 0.8%. We calculated a hospitalisation rate of 1‰ for women having given birth to a child in the Lausanne sector in the years 1958–1977. The mean age of the 57 patients investigated was 26.6 years (range 19–40 years), and all patients were married at the time of confinement. Of the patients 41 (72%) were primiparae, 2 (4%) had completed university studies, 32 (56%) an apprenticeship or a higher professional training and 23 (40%) had no professional training.

Clinical characteristics of the post-partum episode are indicated in Table 2. There were 35% of patients diagnosed as purely depressive and 17% as depressive with schizophrenic symptoms. A remarkable proportion of patients (23%) showed manic syndromes, but this was pure mania only in 2 cases (4%); these 2 patients were added to the group of mania with schizophrenic symptoms. The group of patients with nonaffective psychotic episodes constituted only 25% of the cases. According to the RDC, 5 patients in this group had the diagnosis of an episode of schizophrenia, definite, 6 of schizophrenia, probable, and 3 of unspecific functional psychosis; 1 patient suffering from an infectious toxic psychosis was included in this last-mentioned subgroup. If the RDC had been used in their unmodified form, the principal difference would have been that 5 patients with an episode of mania with schizophrenic symptoms and 1 patient with depression and schizo-

Table 2. Clinical characteristics of post-partum episode of illness (%)

	Total group	Depression	Mania with schizophrenic symptoms, pure mania	Depression with schizophrenic symptoms	Nonaffective psychotic episode
	[$n=57$]	[$n=20$ (35%)]	[$n=13$ (23%)]	[$n=10$ (17%)]	[$n=14$ (25%)]
Delusions	59	15	84	100	71
Hallucinations	45	5	70	80	50
Confusional-oneiroid syndrome	28	0	61	30	35
Onset of illness[a] within 2 weeks after confinement	57	25	89	40	92
Abrupt onset of illness (hospitalisation within 2 weeks after start of illness)	64	35	84	60	92
Duration of illness[b] 6 weeks or less	21	0	15	0	71
Duration of illness 3 months or less	53	30	77	20	86
Patients with "psychic stress" during pregnancy and puerperium	47	70	54	10	36

[a] Defined as the moment when the patient had important symptoms which interfered markedly with her daily activities

[b] Defined as the time elapsed until social remission occurred

phrenic symptoms would have been classified as having a purely affective disorder.

The investigation of some of the principal psychopathological phenomena revealed that hallucinations and delusions were rare in pure depressives and pure manics, but common in all other diagnostic subgroups. Frequently described were delusional or hallucinatory phenomena with some connection to the child or childbirth, without major evidence of inappropriate affect. If a patient had at least three among the following four symptoms: delusions as defined above, hallucinations as defined above, evidence of temporal or spatial disorientation, and marked incoherence, then she was considered as having a confusional-oneiroid syndrome; this was particularly frequent in patients with manic symptoms.

In the majority of cases, the post-partum illness started within 2 weeks after confinement; however, patients with a depressive syndrome tended to become ill later on. An abrupt onset of illness was observed in almost all patients with the appearance of the index episode within 2 weeks after confinement, though this was rarely seen in cases with a late onset of illness. More than half of the patients with a nonaffective psychotic episode had recovered within 6 weeks. This was not the case in the other diagnostic subgroups. In particular, patients with pure depressions and depressions with schizophrenic symptoms tended to have a protracted course of illness.

All the patients' histories were examined for the occurrence of major life stresses which might have played a role in provoking the post-partum illness. The overall rate of patients with a "psychic stress" factor was 47%, the most frequent being chronic marital disharmony (16 cases, 28%), followed by initial ambivalence or a negative attitude towards pregnancy (a total of 11 cases, 20%).

The mean duration of hospitalisation (including rehospitalisations for the index episode) was 64 days. All patients finally left the hospital—the longest hospitalization lasted 7 months—and all improved, at least partially. However, in 8 cases (14%), the post-partum episode led directly to invalidity, either because of only partial remission, or because of very frequent relapses.

Nonpuerperal Relapses

Table 3 shows that 37 patients, i.e., 65%, had nonpuerperal relapses, the rate being 90% in the subgroup of depression with schizophrenic symptoms, and only 43% in patients with nonaffective psychotic episodes. Affective psychosis was the most frequent long-term diagnosis (16 cases, 43%), unipolar endogenous depression being found more often than bipolar manic depressive psychosis (27% compared to 16%). As many as 14 patients (38%) were classified as suffering from schizoaffective psychosis, but only 7 (19%) from schizophrenia.

The rate of 65% of patients with nonpuerperal relapses is high, but this does not mean that all these patients had a poor overall outcome. For example, 5 patients had a single relapse after 10 years or more with complete well being in between. Only 38% of all patients had relapses in the first 5 years after the post-partum episode. Table 4 summarizes the global clinical outcome of all 57 patients. All patients without any further nonpuerperal psychopathology were classified as having a favorable outcome; patients with relapses no more frequent than once every 5 years on average and/or minor episodic or chronic psychopathology as having a relatively favorable outcome; all cases with more frequent relapses and/or other marked long-lasting psychopathology as an unfavorable outcome. Most patients in the last-mentioned group either had areas of life which remained partially intact, and/or they were well between episodes, so it is not justified to speak globally of a poor outcome. Only 25% of patients remained free of any further psychopathology. There were 42% in the intermediate group and 33% had an unfavorable outcome. This group includes the 5 patients who committed suicide; all had frequent relapses without full remission.

Table 3. Proportion of patients with nonpuerperal relapses and long-term diagnoses in relation to diagnostic categories of the post-partum episode of illness

	Total group	Depression	Mania with schizophrenic symptoms, pure mania	Depression with schizophrenic symptoms	Nonaffective psychotic episode
	($n=57$)	($n=20$)	($n=13$)	($n=10$)	($n=14$)
Total of relapses (percentage values)	65	60	77	90	43
Subgroup of 37 patients with psychotic nonpuerperal relapses (absolute values)					
Unipolar endogenous depression	10	7	-	1	2
Bipolar manic depressive psychosis	6	3	3	-	-
Schizoaffective psychosis	14	1	7	6	-
Schizophrenia	7	1	-	2	4

To describe the course of illness in the diagnostic groups according to long-term classification, we evaluated two characteristics: the degree of remission between the episodes of illness, in the 5 years preceding the follow-up investigation, and the mean cycle length per patient, i.e., the mean interval between the beginning of one episode and the next one. In a few cases with frequent relapses, the best possible estimate was made. To calculate the cycle length, we excluded periods of full remission under lithium treatment. Patients with unipolar endogenous depression had relatively few episodes, the cycle length being 6.1 years, as compared to 2.7 and 3.1 for patients with bipolar manic depressive psychosis and schizoaffective psychosis respectively. Three patients with schizoaffective psychosis had an unipolar manic course of illness, 7 a bipolar one, and 4 a unipolar depressive one. Here we found the highest percentage of cases with residual symptoms between episodes of illness (50%) compared with 20% and 18% for unipolar depression and bipolar manic depressive psychosis. Among the patients with the long-term diagnosis of schizophrenia, 3 developed severe chronic schizophrenia. The 4 other patients had sporadic episodes, with full remission in 3 cases. Also in the group of patients without nonpuerperal relapse, a fourth showed psychopathology in later evolution.

A change of symptomatology comparing the post-partum episode with nonpuerperal relapses was observed in a number of cases (see also Table 3). Among the 25 patients who had a diagnosis other than pure depression or mania at the post-partum episode and who had nonpuerperal relapses later, 5 went exclusively through episodes with pure affective symptoms. Furthermore, confusional elements were definitely less frequent in nonpuerperal relapses. The later appearance of schizophrenic symptoms in patients with pure affective disorders at the post-partum episode was observed twice.

Puerperal Relapses

There were 26 women who had a total of 32 further childbirths. In 11 cases (34%), a further severe psychic decompensation occurred. In none of the 11 cases was the long-term course of the illness negatively influenced by this additional puerperal decompensation. However, 2 patients with particularly unfavorable evolutions had slight puerperal episodes before the index episode, after which the chronic, invalidating evolution began.

Information on Marital Status and Evolution of the Child

In total 84% of marriages continued until the follow-up, or until the death of the patient (4 cases) or the husband (2 cases). For the previous 5 years, two-thirds of these relations had been generally harmonious and one-third chronically conflictual; 16% of the marriages ended in divorce. All but 4 patients took care personally of the child born at the index episode; prolonged placements with grandparents or other relatives at periods other than relapses were not observed. A screening of the evolution of the children revealed that the majority of the 54 children, on whom concrete information was available, did well and caused no particular problems. No case came to our knowledge during the follow-up period of any child who had been physically injured by the patient for psychotic or other reasons. Some psychopathological phenomena were seen in 19 children (i.e. 35%), at least once during childhood or puberty; contact difficulties and behavior disorders were the most frequent.

Treatment Received During Follow-up

After the post-partum episode, the patients as a whole received a considerable amount of psychiatric care. All but 2 of the 37 patients with nonpuerperal relapses had been rehospitalised at least once; 8 patients five times or more; no patient, however, remained permanently in an institution. At follow-up, 24 patients (42%) were under medical treatment for psychic troubles, the majority of them with psychiatrists; 11 patients were taking lithium. In 7 cases, including 3 with schizoaffective psychosis, lithium has led to full remission, as can be deduced from the absence of further episodes after former frequent relapses.

Psychopathology Before the Index Episode, Family History of Psychosis

For psychopathology before the index episode, two groups were distinguished: psychotic episodes (not leading to hospitalisation), and nonpsychotic psychopathology. Seven patients had had psychotic episodes, and 8 patients had nonpsychotic diagnoses. Not included in either group were mild puerperal decompensations before the index episode, which occurred in a few cases.

A total of 15 patients (26%) had one or more first degree relatives with psychoses. Hereditary antecedents were found mainly in women with affective and schizoaffective psychoses. In the group of schizophrenia, there was just 1 case, and none among the patients without nonpuerperal relapse. In the majority of patients with family histories of psychosis, affected relatives suffered from affective psychosis, but 3 cases with a first degree relative with schizoaffective psychosis and 3 cases with a first degree relative with schizophrenia were also found.

Characteristics Related to Puerperal and Nonpuerperal Relapse and Unfavorable Global Outcome

The frequency of 27 variables, including background data, family history of psychosis, psychopathological characteristics and diagnoses of the index episode, were compared in patients with and without nonpuerperal relapse. Only the 3 following criteria were significantly related to relapse (χ^2): (1) family history of psychosis ($P<0.001$), (2) psychotic episodes before the index one ($P<0.05$), and (3) duration of the post-partum episode of more than 6 weeks ($P<0.01$). All patients with the first and second characteristic had nonpuerperal relapses, but a number of patients with long duration of the index episode had none. This criterion is less reliable since the duration of the illness might have been influenced by the choice of treatment. No relation to relapse was found for any of our diagnoses at the post-partum episode, nor for the RDC diagnosis of a schizophrenic episode.

An analogous statistical evaluation for risk factors for puerperal relapses was not possible because of the small size of the groups; however, there was one clearcut finding: only 1 puerperal relapse occurred in the total of 11 childbirths in patients without nonpuerperal relapse, in comparison to 10 relapses among 21 childbirths in patients with nonpuerperal relapse, the difference being statistically significant ($P<0.05$).

The 3 characteristics significantly related to an unfavorable global outcome were: (1) any psychopathology (psychotic or nonpsychotic) before the index episode ($P<0.05$), (2) a diagnosis of depression with schizophrenic symptoms at the index episode ($P<0.05$), and (3) an onset of illness later than 2 weeks after confinement ($P<0.05$).

Discussion

General Characteristics of the Sample, Psychopathology of Post-Partum Episode of Illness

The selection criteria of the 57 patients described differ in one respect from all other follow-up studies we know of: only patients in whom the puerperal disorder was the first major psychic decompensation leading to hospitalisation were included. These women therefore represent a particularly pure sample of cases with childbirth related onset of unique or recurrent psychopathology. This selection criterion is of importance when a comparison is made between the clinical characteristics of patients with a puerperal onset of psychotic illness and those assigned to the corresponding diagnostic subgroups in general.

The hospitalisation rate of 1.1 per 1000 confinements calculated for the present sample is at the lower limit of the frequently reported rate of 0.1% to 0.2%; this indicates that we are dealing principally with severe decompensations. The results obtained should not be extended to the large group of nonhospitalised, nonendogenous depressives. In this group long-term studies are lacking and the puerperal episode of illness might possibly be the only one in the patient's life.

The 57 patients showed a number of well known characteristics at the post-partum episode, like the predominance of primiparae (Kendell et al. 1981), the almost total absence of severe physical complications of the puerperium, and the abrupt beginning of the illness, within 2 weeks after delivery in a substantial proportion of cases (Brockington et al. 1978). The finding that all patients were married at the time of confinement is unusual and at variance with investigations reporting an association between illegitimacy and puerperal disorders (Tetlow 1955; Jansson 1964; Kendell et al. 1981), but at the same time it corresponds to the findings of other studies describing a high proportion of married women (Saeger 1960; Paffenbarger 1964; Osterman 1963; Protheroe 1969; Da Silva and Johnstone 1981). The distribution of diagnoses for the post-partum episode of illness can be compared with three investigations in which the RDC were used. Dean and Kendell (1981) and Katona (1982), who made retrospective studies, used very similar inclusion criteria, whereas Brockington et al. (1981), in a prospective study, included only patients with onset of illness within the first 2 weeks after childbirth. Our investigation is in agreement with the results of all three of these studies concerning the finding that pure depression was the most frequent diagnosis, that there was quite a high proportion of cases with a manic syndrome and that nonaffective psychotic episodes were rather rare. Not only with the wide definition for episodes of illness with affective and schizophrenic symptoms we used in our study, was their frequency particularly high. Brockington et al. (1981), using the unmodified RDC, found several cases with schizoaffective manic episodes which were over-represented in comparison with controls; however, schizoaffective depressive episodes were under-represented. Da Silva and Johnstone (1981), in their sample, made a diagnosis of puerperal schizophrenia in 40% of their cases; however, the difference is presumably due to the diagnostic concept (Present State Examination) (Wing et al. 1974), which includes patients with a full affective syndrome.

Proportion of Patients with Nonpuerperal Relapses

To our knowledge, the proportion of 65% of patients with nonpuerperal relapses and of 75% with psychopathology of any type during follow-up found in the present investigation is the highest so far reported in the literature. Arentsen (1968) found nonpuerperal psychotic relapses in 35% and nonpuerperal psychotic or nonpsychotic psychopathology in 61%, and these proportions increase to 44% and 67% respectively, if cases with chronic psychotic evolution are included. In Protheroe's investigation (1969), 39% of patients had nonpuerperal psychotic relapses, but the observation period was short in a number of cases. We have already mentioned that the proportions found in the other investigations listed in Table 1 cannot be considered as indicating the real risk because of the insufficient observation time. However, other factors must also be responsible for the variation in the proportion of patients with nonpuerperal relapses in the different investigations. First, there are possible differences in the use of the term "psychotic relapse". The higher proportion of patients with psychotic episodes unrelated to childbirth among all nonpuerperal psychopathological phenomena during follow-up in our investigation compared to Arentsen's study might indicate that we have used a larger concept of psychosis. Secondly, a presumably important factor lies in the variation in local attitudes toward

the admission or nonadmission to psychiatric hospitals of patients with moderate or very brief decompensations—cases which are largely absent in our study. Thirdly, the samples catamnestically investigated in former investigations were slightly different in that toxic organic psychoses were somewhat more frequent and that a number of patients with "puerperal schizophrenia" (Protheroe) and "delirium acutum" (Arentsen) died in the course of the index episode. Finally, possible lack of complete information caused by the manner of collecting data might have led to a too optimistic prognostic outlook. Martin (1958), who reported the most favorable long-term evolutions, used a mailed questionnaire as his principal instrument.

For the foregoing reasons, the different investigations are not completely comparable. Nevertheless, the results of the present investigation can be considered as a confirmation of Protheroe's statement (1969) that after a long enough observation time, more than half of the patients with severe post-partum disorders will have nonpuerperal relapses. Our results are furthermore in agreement with those of Da Silva and Johnstone (1981), who consider that it is rather rare for a woman with a severe post-partum disorder to become symptom free within 1 to 6 years, and complete them by the finding that psychopathology is likely to occur also in later evolution.

Our results show some general progress in psychiatric treatment. No patient was chronically hospitalised and several recovered wholly or partially because of the long-term treatment they received. In addition, psychotherapeutic measures helped definitely in improving social functioning.

Clinical Characteristics of Endogenous Psychoses with Puerperal Onset

So far, little attention has been paid to the detailed diagnostic and clinical characterization in general of the later nonpuerperal course of illness. Foundeur et al. (1957) considered that the majority of patients with nonpuerperal relapses were suffering from schizophrenia, whereas Arentsen (1968) believed that two-thirds suffered from affective psychosis. In Da Silva and Johnstone's investigation (1981), the relation of diagnoses of affective psychosis to schizophrenia was 2 : 1 at follow-up. Foundeur et al. (1957) came to the conclusion that puerperal and nonpuerperal schizophrenia followed an identical course. Almost all patients classified by Arentsen (1968) as schizophrenic had a chronic course of illness, whereas all patients with affective psychosis had an episodic course with remissions. Protheroe (1969) and Da Silva and Johnstone (1981) found similar results and noted the poorer prognosis of schizophrenia with puerperal onset as compared with affective psychosis. Huhn and Drenk (1973), on the basis of an incomplete follow-up investigation, concluded that 80% of patients with later nonpuerperal episodes of illness belonged diagnostically to affective psychosis. Indirect information on diagnostic classification of disorders starting or reappearing in the puerperium can be obtained from investigations in which the influence of childbirth on the course of endogenous psychoses was studied. Bratfos and Haug (1966) and Reich and Winokur (1970) found that more than 20% of confinements of patients with affective psychosis were followed by episodes of illness. To our knowledge, no such study exists for schizoaffective psychosis, but Tsuang et al. (1976), in an investigation of "atypical schizophrenia",—the clinical characteristics of the sample corresponding in part to our definition of schizoaffective psychosis—found a higher childbirth-related onset of illness than in affective psychosis or schizophrenia. Also "cycloid psychoses" (Leonhard 1979; Perris 1974), which overlap with schizoaffective psychoses but also include some recurrent nonaffective psychoses usually classified as schizophrenia, were found to be prone to puerperal decompensations. Perris (1974) reported that 14 of 21 childbirths observed in a group of 44 women were followed by psychotic episodes. Boeters (1971), in a group of 45 women with "oneiroid-emotion psychoses"—a diagnostic concept similar to schizoaffective and cycloid psychoses—found puerperal decompensations in 31% of the confinements. The association between atypical psychoses and puerperal psychoses has also been claimed by other authors (Pauleikhoff 1964; Janssen and Denker 1964; Grosse 1968; Mentzos 1968). In opposition to this, the course of schizophrenia—apart from the rare cycloid psychoses—is little influenced by childbirth, as demonstrated by Yarden et al. (1966).

The studies mentioned suggest that affective psychosis and schizoaffective psychosis might be very frequent among childbirth-related psychoses. In our investigation, the high proportion of cases with schizoaffective psychosis is striking (Table 3). The following calculation will show that the distribution of diagnoses found in our sample differs from the incidence of endogenous psychoses in a general population of women, aged 20–40 years. We take the estimate often found in the literature that the lifetime risk of schizophrenia in the general population is 1%, and take the estimated rate of 1.5% for affective psychosis (Odegard 1972) with a relation of about 2 : 1 between unipolar depression and bipolar manic depressive psychosis, calculated from a study of Angst (1980) excluding cases of unipolar depression with a single episode. If we assume that schizoaffective psychosis is no more than one-third as frequent as schizophrenia and that half of the cases with schizoaffective psychosis are included in the 1% with schizophrenia and half in the 1.5% with affective psychosis, we arrive at the distribution of lifetime risks indicated in Table 5. Next, we consider sex ratios and the proportion of cases with onset of illness between 20 and 40 years for endogenous psychoses, referring to standard indications in the literature (Bleuler 1972; Perris 1981) and respectively in an investigation of Angst (1980) in which similar diagnostic criteria were used. From these data we arrive at the calculated distribution of the incidence of endogenous psychoses (Table 5) demonstrating the very high representation of schizoaffective psychosis in our sample. This principal finding seems certain, even if the calculation may be open to some criticism for its use of estimated values or by a degree of arbitrary choice.

The rather low number of patients with pure affective psychosis in our investigation is surprising in view of the aforementioned results of a high frequency of puerperal episodes in patients with this diagnosis (Bratfos and Haug 1966; Reich and Winokur 1970). It is possible that a greater proportion of patients with affective psychosis, particularly with pure depression receive only outpatient treatment than is the case in patients with schizophrenic symptoms. Considering this point, our conclusion is that our results clearly indicate that schizoaffective psychosis begins much more frequently in the puerperium than does schizophrenia, compared with the incidence of endogenous psychoses of the corresponding age group in women, but it is not certain whether it also begins more frequently than affective psychosis.

Another question to be clarified is whether psychoses with puerperal onset have atypical characteristics in comparison to

Table 4. Global clinical outcome and psychic health at follow-up (%)

Outcome	Long-term diagnosis					
	Total group	Unipolar endogenous depression	Bipolar manic depressive psychosis	Schizo-affective psychosis	Schizo-phrenia	Patients without non-puerperal relapses
	($n=57$)	($n=10$)	($n=6$)	($n=14$)	($n=7$)	($n=20$)
Global clinical outcome: favorable / relat. favorable / unfavorable	25/42/33	0/50/50	0/83/17	0/43/57	0/57/43	70/20/10
Psychic health at follow-up: well / slight psychopathological symptoms / marked psychopathology / dead	58/19/14/9	50/30/10/10	83/0/17/0	36/29/14/21	43/0/43/14	75/20/5/0

Table 5. Calculated incidence of endogenous psychoses in women aged 20–40 years: comparison of distribution of diagnoses with patients suffering from a severe post-partum disorder

	Unipolar endogenous depression	Bipolar manic depressive psychosis	Schizo-affective psychosis	Schizo-phrenia
Lifetime risk (%)	0.91	0.45	0.28	0.86
Percentage of women in diagnostic subgroup	75[a]	61[a]	73[a]	50[b]
Percentage of patients with onset of illness between 20 and 40 years	32[a]	50[a]	59[a]	67[c]
Calculated incidence in women, aged 20–40 years	0.22	0.13	0.12	0.29
Calculated distribution of diagnoses in % (incidence in women, aged 20–40 years)	29	17	16	38
Distribution of diagnoses in 37 patients with severe post-partum disorders	27	16	38	19

[a] Angst, 1980
[b] Bleuler, 1972
[c] Perris, 1981

the usual course of illness of the corresponding diagnostic subgroup. For comparison of affective and schizoaffective psychosis, we refer to the investigation of Angst (1980). Our evaluation differs essentially from that of Angst on two points, i.e., the exclusion of periods with lithium treatment and the exclusion of patients with a single episode. The author found similar cycle lengths for unipolar depression, bipolar manic depressive psychosis and schizoaffective psychosis (4.2, 2.5 and 2.9 years), and the proportion of patients with partial remission in the three subgroups is also comparable. Also the small group of patients with schizophrenia in our investigation did not show major evidence of unusual characteristics in long-term evolution.

Kadrmas et al. (1979) found that patients with puerperal onset of manic disorder have a different course of illness from patients with manic onset unrelated to childbirth, the diagnostic concept including patients with mania and schizophrenic symptoms. In patients with puerperal onset the authors found no nonpuerperal relapses in the following 3 years, which was significantly different from controls. Our group of patients with manic syndrome at the index episode and later relapses did not show strong evidence of an unusual course of illness, however there is a partial confirmation by the fact that 3 of the 13 patients had no relapses in later life. This is exceptional in comparison with the usual evolution of bipolar manic depressive psychosis and schizoaffective psychosis (Angst 1980).

The Liability of Schizoaffective Psychosis to Puerperal Decompensations: Its Possible Nosological Significance

The differential susceptibility to puerperal decompensations of different diagnostic subgroups by the "standard stress" of birth indicates some kind of heterogeneity; however the question remains unanswered whether this is a difference associated with the primary pathophysiological disturbances or a rather peripheral difference, such as stress susceptibility. Under the assumption that the first is the case, the difference can help to provide information on pathophysiological entities, even in the presence of different clinical pictures.

Along with other unsolved questions about the nosology of endogenous psychoses, the concept of schizoaffective psychosis is still under discussion. Procci (1976), on the basis of an extensive review of the literature, found that schizoaffective

psychosis is probably a heterogeneous entity, some cases representing a variant of affective psychosis or an independent entity bearing a similarity to affective psychosis, while others are related to schizophrenia, and a few are true mixtures between the two major endogenous psychoses. Recent genetic investigations offered further evidence for one or another of these assumptions, like heterogeneity and possibility of attribution to affective psychosis and schizophrenia respectively (Tsuang 1979), a close relation to affective psychosis in the majority of cases (Mendlewicz et al. 1980), or an independent intermediate position between affective psychosis and schizophrenia (Angst et al. 1979a and 1979b). Some heterogeneity is suggested not only by genetic data, but also by the course of illness and response to treatment; e.g., the subgroup of schizoaffective patients with manic decompensations is similar in this respect to affective psychosis (Abrams and Taylor 1976; Brockington et al. 1980a; Rosenthal et al. 1980). This close relation does not seem to exist for schizoaffective psychosis with a unipolar depressive course (Brockington et al. 1980b).

Our results of a particular and possibly uniquely strong liability of schizoaffective psychosis to a puerperal onset of illness situate this diagnostic category, with respect to this characteristic, close to affective psychosis and separate it from schizophrenia. Furthermore, some homogeneity of schizoaffective psychosis is indicated by the finding that patients with unipolar depressive, bipolar and unipolar manic courses of illness were represented.

Patients with Exclusive Puerperal Decompensations

The question to consider is how patients without nonpuerperal relapses should be diagnostically classified. For two reasons, it is unlikely that pure depressives without relapse are to be considered as being related to affective psychosis: it is infrequent that patients with affective psychosis have a single episode in life (Angst 1980), and there was no family history of affective psychosis in this group. On the other hand, no evident neurotic conflicts were observed which would have justified the diagnosis of neurotic depression. It seems, therefore, correct to consider these depressions as atypical (Pitt 1968). The arguments of an absence of family history of psychosis and the rarity of a course of illness with a single episode also argue against the possibility that the other cases with psychotic puerperal episodes without nonpuerperal relapse are related to one of the traditionally recognized endogenous psychoses. They seem to be real post-partum psychoses in the sense of a separate clinical entity.

Puerperal Coloration of Endogenous Psychoses with Childbirth-Related Onset

The frequently described atypical characteristics of severe post-partum disorders can partially be explained by the high proportion of cases which do not belong to the two major endogenous psychoses. However, there is also a specific puerperal factor involved, as shown by the absence of nonaffective psychotic and confusional symptoms in nonpuerperal relapses as compared to the index episode, in a number of cases. The temporarily increased fragility of the CNS caused by the important endocrinological changes in this period is probably the biological basis for the appearance of these unique puerperal symptoms.

Risk Factors for Nonpuerperal and Puerperal Relapse and Unfavorable Global Outcome

Although many authors have written about the prognosis for severe post-partum disorders, there are few investigations in which criteria related to relapse and clinical outcome in general were evaluated.

The validity of the two criteria strongly related to and exclusively found in our study in patients with nonpuerperal relapse, i.e. heredity of psychosis, and occurrence of psychotic episodes before the index one, seems obvious. Both criteria make it very likely that we are dealing with one of the usual endogenous psychoses, and all endogenous psychoses have a high tendency to recurrences.

Our results, showing that patients with nonpuerperal relapses also have a much higher risk of puerperal relapses than patients without nonpuerperal relapses, are in full agreement with Arentsen's findings (1968). Protheroe (1969) does not comment on this problem. Further evidence of this risk factor is the finding of the high frequency of puerperal episodes in patients with affective, schizoaffective and cycloid psychoses.

Among the criteria related to a globally unfavorable outcome in our study, one of them, the occurrence of psychopathology before the index episode, has already been revealed as a risk factor by other investigators (Jansson 1964; Wilson et al. 1972). Little can be said about the validity of the criterion of the beginning of post-partum illness later than 2 weeks after confinement, the latter being the only one found in our study. Also the third criterion, a diagnosis of depression with schizophrenic symptoms in the post-partum episode, is difficult to interpret. It might partially correspond to the finding of a number of authors on the negative prognostic importance of a diagnosis of puerperal schizophrenia (Hemphill 1952; Protheroe 1969; Da Silva and Johnstone 1981). It should be mentioned, however, that we did not find a relation between a negative outcome and schizophrenia as a long-term diagnosis. Our principal finding was the rarity of the diagnosis.

Finally, we found some possible indications of a chronic deterioration of psychic health as a consequence of repeated puerperal decompensations. Protheroe has clearly described such cases. Therefore, termination of pregnancy may be justified for medical reasons. However, having had a puerperal disorder is certainly not in itself a sufficient reason for termination of pregnancy or for definite advice to have no further children. Women should be correctly informed about the risk but, with rare exceptions, they should not be discouraged from having another child if they strongly desire it.

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