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Catatonia in the peripartum: A cohort study using electronic health records

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ABSTRACT

Background: Due to limited existing literature available on the presentation and treatment of catatonia in the peripartum, this retrospective descriptive cohort study aimed to examine demographic data, catatonic features, diagnoses pre- and post-catatonic episodes, treatment and the presence of obstetric complications.

Methods: Individuals with catatonia were identified in a previous study using anonymised electronic healthcare records from a large mental health trust in South-East London. The presence of features from the Bush-Francis Catatonia Screening Instrument was coded by the investigators and longitudinal data were extracted from structured fields and free text.

Results: 21 individuals were identified from the larger cohort, each of whom experienced one episode of catatonia in the postpartum period, and all had had an inpatient psychiatric admission. 13 patients (62 %) presented after their first pregnancy and 12 (57 %) experienced obstetric complications. 11 (53 %) attempted breastfeeding and 10 (48 %) received a diagnosis of a depressive disorder following the episode of catatonia. The majority presented with immobility or stupor, mutism, staring and withdrawal. All were treated with antipsychotics and 19 (90 %) received benzodiazepines.

Conclusions: This study suggests that signs and symptoms of catatonia during the peripartum are similar to other catatonic presentations. However, the postpartum may be a period of high risk for catatonia and obstetric factors, such as birth complications, may be relevant.

1. Introduction

Catatonia is a neuropsychiatric disorder that typically presents with a combination of behavioural and motor signs (Kahlbaum, 1874). Clinical manifestations include stupor, mutism, waxy flexibility, unusual postures and staring (Farias and Hartnett, 2022). Catatonia is commonly described as having three main clinical subtypes: "excited" with excessive and purposeless motor activity, "stuporous" with mutism, inhibited movement, staring and negativism and "malignant" which is a life-threatening state that presents with fever, autonomic instability, delirium and rigidity (Csihi et al., 2022). The aetiology is complex and it can occur in mental disorders such as severe mood disorders or schizophrenia. Catatonia can also be seen in those with neurological conditions (for example meningitis and autoimmune encephalitis) and other infectious, metabolic or rheumatological disorders (Farias and Hartnett, 2022). The prevalence of catatonia in those with an acute mental disorder is reported to be between 7 % and 17 % (Solmi et al., 2017).

Pregnancy and the postpartum can be an emotionally and physically challenging time. This may be due to hormonal changes, sleep deprivation, psychosocial stressors, obstetric complications, traumatic birth and inadequate social support. Mental disorders are the commonest complication of childbearing and are associated with considerable maternal and infant morbidity and mortality (Howard and Khalifeh, 2020). The early postpartum is associated with a high risk for new and recurrent episodes of severe mental illness, with around one in 1000 requiring admission to a psychiatric hospital in the first few months following birth (Jones et al., 2014). Lisette and Crystal (2018) found that approximately one in 13 individuals who are pregnant develop a major depressive episode. Di Florio et al. (2021) recently found that some individuals have a heightened genetic vulnerability to postpartum illness, even if they have had no previous psychiatric history, particularly in Bipolar Affective Disorder, which may manifest for the first time in the

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postnatal period as postpartum psychosis.

Although there is a growing body of research on the psychopathology of mental disorders in the peripartum, there is limited literature available on the presentation and treatment of catatonia in the peripartum. The existing literature is limited to single case reports and one small case series (Csihi et al., 2022), but further research is necessary to understand the role of catatonia in peripartum mental disorders. This study aimed to characterise the presentation of catatonia in the peripartum period in a South-East London cohort.

2. Methods and participants

2.1. Study design

A retrospective descriptive cohort study was conducted to examine the demographic, obstetric and clinical aspects of peripartum catatonia.

2.2. Setting

South London and Maudsley NHS Foundation Trust (SLaM) provides NHS mental health services to one of the most ethnically diverse populations in the UK. SLaM provides inpatient and community services to 1.3 million people in South London in addition to specialist services to adults across the UK. Perinatal mental health services provide care to those with a mental disorder who are either pregnant or in the first year postpartum. Perinatal services include specialist community perinatal mental health teams serving the local boroughs of Lambeth, Southwark, Lewisham and Croydon and a 13-bed inpatient Mother and Baby Unit, which is a regional and national unit.

2.3. Participants

The Clinical Record Interactive Search (CRIS) system comprises anonymised mental healthcare records from approximately 400,000 individuals with a history of mental disorder seen in community and inpatient settings and emergency departments in SLaM. The system comprises all mental healthcare records from 2006 onwards and some from as early as 1999 (Stewart et al., 2009). The CRIS system has ethical approval from the Oxfordshire C Research Ethics Committee (ref: 18/SC/0372) and the current study was approved by the CRIS Oversight Committee (ref: 17-102).

In an original study by Rogers et al. (2021), CRIS was searched for all mentions of 'catatonia' or 'catatonic' in patients' notes up to 17 December 2018, including all inpatients and outpatients. A natural language processing application was used to remove obviously irrelevant entries, then researchers manually viewed each record to ensure eligibility. Included episodes needed to have a diagnosis of catatonia made by a clinician according to their clinical judgement, at least two clinical features from the Bush-Francis Catatonia Screening Instrument evident at the time of this diagnosis (Bush et al., 1996) and an identifiable index date for the catatonia. Among the 1456 individuals identified in this study, our cohort was defined as those who were being treated by a perinatal mental health team at the time of onset of catatonia. Under the current service configuration, all perinatal patients with a mental illness of moderate or severe intensity are cared for by a community or inpatient perinatal mental health team, so it is unlikely that any patients with catatonia in the perinatal period would be missed with this search strategy.

2.4. Data extraction

Age, ethnicity, clinical diagnoses and admission duration were extracted directly from the structured fields of the patient record. Parity, breastfeeding status, individual catatonic signs, duration of catatonia, timing relative to birth, treatment of catatonia and treatment complications were manually extracted from the patient record by the first author.

3. Results

21 individuals were identified, each of whom experienced one episode of peripartum catatonia. All developed catatonia in the postpartum and all underwent inpatient psychiatric admission. The demographic characteristics of the group are outlined in Table 1. Mean age was 32 years with 52 % belonging to a Black/African/Caribbean/Black British background. 5 individuals had a significant past medical history, consisting of a neurological disorder ($n = 1$), a chronic respiratory disease (1) and an endocrine disorder (3). 12 individuals had experienced obstetric complications. These complications included hypothyroidism during pregnancy, gestational diabetes, pre-eclampsia, traumatic forceps deliveries, postpartum haemorrhage requiring blood transfusion, meconium aspiration requiring emergency Caesarean section, third-degree tear and Group B Streptococcus infection. 11 patients attempted breastfeeding but three had to discontinue, either due to medication-related drowsiness or increasing agitation.

Characteristics of the catatonic episodes are shown in Table 2. The most common diagnoses made prior to the onset of catatonia were major depression ($n = 5$) and severe mental and behavioural disorder associated with the puerperium (5). The most common diagnosis made after the onset of catatonia was major depression (10), of whom 9 had psychotic features. Antipsychotic medication was used in all patients. Benzodiazepines were used in 19, of whom 18 used lorazepam, eight clonazepam and two diazepam (some individuals using more than one agent from this class).

The frequencies of specific catatonic features are illustrated in Fig. 1. The majority of the patients presented with immobility or stupor, mutism, staring and withdrawal.

4. Discussion

4.1. Findings in context

This study identified 21 patients from an ethnically diverse population who presented with catatonia in the postpartum period. 62 % presented after their first pregnancy and 57 % experienced obstetric complications. All of them required an inpatient psychiatric admission for the management of their symptoms.

Perhaps the most striking finding is that no episodes of catatonia were identified during pregnancy, suggesting an increased vulnerability in the postpartum relative to the antepartum period. The postpartum is already thought to be a time of increased vulnerability to an episode of severe mental disorder relative to the antepartum (Nguyen et al., 2022). However, the distribution of catatonic features among the perinatal patients in this study is very similar to that in the wider cohort of 1456 patients with catatonia, from which this perinatal sample was drawn (Dawkins et al., 2022). This echoes the findings of a review comprising two small retrospective case series and 20 case reports, which suggested that perinatal catatonia is similar in presentation to catatonia in other

Table 1
Demographic and obstetric characteristics.

Variable	Properties ($N = 21$)
Age (mean, SD)	32 (7.2)
Ethnicity (n, %)	
- White	8 (38)
- Asian	2 (10)
- Black	11 (52)
Parity (n, %)	
- 1	13 (62)
- 2–5	8 (38)
Obstetric complications (n, %)	12 (57)
Breastfeeding at least partially (n, %)	11 (53)

Table 2
Clinical characteristics.

Variable ($N = 21$ unless otherwise stated in footnotes)	Properties
Treatment setting, *n* (%)	
- Mother and baby unit (MBU)	17 (81)
- Perinatal mental health team	2 (10)
- Other acute psychiatric wards	2 (10)
Time postpartum of catatonia onset (days), mean (SD)[a]	120.2 (116.8)
Time postpartum of catatonia onset (days), median (IQR)[a]	78.5 (200.5)
Duration of catatonia (days), mean (SD)[b]	23.4 (32.1)
Duration of catatonia (days), median (IQR)[b]	9 (23.5)
Number of BFCSI features, median (IQR)	4(4)
Hallucinations (*n*, %)	
- Any	13 (62)
- Auditory	13 (62)
- Visual	5 (24)
Delusions (*n*, %)	
- Any	16 (76)
- Grandiosity	2 (10)
- Paranoia	15 (71)
- Nihilism	4 (19)
- Passivity	2 (10)
Suicidal ideation (*n*, %)	6 (29)
Suicide attempt (*n*, %)	0 (0)
NCS, median (IQR)	
- Motor subscale	1 (1)
- Affective subscale	6 (2)
- Behavioural subscale	3 (1)
- Total	11 (4)
Diagnosis prior to catatonia onset (*n*, %)	
- Bipolar affective disorder	2 (13)
- Major depression	5 (33)
- Severe mental and behavioural disorder associated with the puerperium	5 (33)
- Schizophrenia spectrum disorder	2 (13)
- Not specified	1 (7)
Diagnosis after catatonia onset (*n*, %)	
- Major depression	10 (48)
- Schizophrenia spectrum disorder	4 (19)
- Severe mental and behavioural disorder associated with the puerperium	5 (23.8)
- Not specified	2 (10)
Treatments used (*n*, %)	
- Antidepressant	13 (62)
- Antipsychotic	21 (100)
- Benzodiazepine	19 (90)
- Electroconvulsive therapy (ECT)	2 (10)

BFCSI – Bush-Francis Catatonia Screening Instrument. NCS – Northoff Catatonia Scale.
[a] $N = 12$.
[b] $N = 19$.

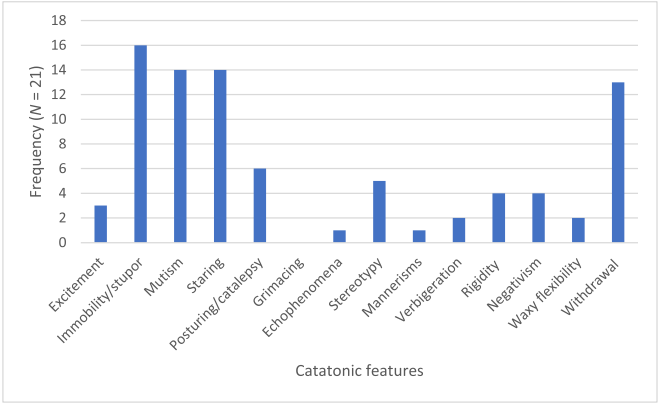

Fig. 1. Number of patients with specific catatonic features from the Bush-Francis Catatonia Screening Instrument.

contexts (Csihi et al., 2022). However, it is likely that in our samples (both the smaller perinatal one and the larger, more inclusive one) under-recognition of certain clinical signs may have played a role, although the distribution of clinical features is not dissimilar from Bush et al.'s prospective study of patients with catatonia (Bush et al., 1996).

The finding that the catatonic features in this cohort are similar to those observed in the general, non-perinatal population echo the findings of two small retrospective case series and 20 case reports, which suggested that perinatal catatonia is similar in presentation to catatonia in other contexts (Csihi et al., 2022).

Many cases (52 %) were from a Black/African/Caribbean/Black British background. Several existing studies suggest higher rates of catatonia in the Black population (Hutchinson et al., 1999; Rogers et al., 2021). To our knowledge, this study is the first to report this in the perinatal population. Ethnic differences could be due to differing interpretations of symptoms by clinicians of the dominant culture (Hutchinson et al., 1999) or as a result of schizophrenia being more common among migrant populations (Rusner et al., 2016). However, many in the cohort received a diagnosis of depressive disorder, with 90 % of these being diagnosed with psychotic depression.

Treatment varied between individuals, although all those in the study received antipsychotics. Only 10 % of the cases received ECT for treatment of catatonia. The presence of catatonia and suicidality in postpartum psychosis are common indications for treatment with ECT (Babu et al., 2013). Therefore the rationale for ECT use being uncommon in this sample is unclear. It si possible that stigma around the use of ECT may have been a factor.

Approximately half of individuals were able to breastfeed at least partially despite being seriously unwell. Indeed existing literature suggests that maternal schizophrenia is associated with a lower likelihood of breastfeeding initiation post-delivery (Taylor et al., 2021; Hill et al., 2019) but that there is still usually a desire to breastfeed (Baker et al., 2021). The literature on the safety of many psychotropic drugs in lactation is sparse and often limited by residual confounding (McAllister-Williams et al., 2017). The current evidence suggests that second-generation antipsychotics are relatively safe in the short term, but the available data are largely for olanzapine and eschew longer-term outcomes (Uguz, 2016). In terms of benzodiazepines, the relative infant dose in one small study was less than 10 % in lactation for most agents (Nishimura et al., 2021), but the safety of using benzodiazepines at the high doses sometimes required in catatonia remains uncertain.

4.2. Strengths and limitations

This study is one of the largest examining catatonia in the perinatal period. It is thus at substantially lower risk of reporting bias than the many case reports on the subject. Moreover, the combination of coded data in structured fields with manual review of free text allows a high degree of detail about the included subjects.

However, this study has a number of limitations. Firstly, this is a retrospective study that relies on documentation of clinical details, some of which may have been missed. The study relies on the judgement of a clinician in correctly identifying and recording catatonic signs as well as duration of catatonia and response to treatment. Although our study is more inclusive than some others relying on electronic healthcare records, which have used formal discharge diagnoses, it still required positive identification of catatonia, which may underestimate the true prevalence of the condition. From review of clinical notes, information on duration of catatonic symptoms, severity of symptoms and impact of treatment, including impact on the infant, were often vague. This may have led to inaccurate notes and clinical records being incorrectly excluded.

5. Conclusions

Future research may usefully examine aspects that are potentially

unique to the peripartum such as duration of catatonic symptoms or response to treatment. It is not clear whether the presence of catatonia in the peripartum is associated with poorer prognosis or longer hospital admissions as there are no studies in the current literature examining this. A transcultural prospective perinatal cohort conducted internationally could address this evidence gap.

It is well established that severe mental illness is associated with an increased risk for adverse obstetric outcomes including preterm birth, foetal growth restriction, pre-eclampsia, antepartum and postpartum haemorrhage, placental abruption and stillbirths (Vigod et al., 2015; McAllister-Williams et al., 2017; Stein et al., 2014). The presence of catatonic symptoms can exert an additional burden on maternal and infant health. For example, immobility and poor oral intake can lead to physical health complications such as dehydration and increased risk of complications such as venous thromboembolism. Furthermore, the presence of catatonia may have a significant impact on ability to provide infant care, including practical care such as feeding but also emotional care, which can have implications for attachment (Csihi et al., 2022). Therefore further research on the aetiology and management of peripartum catatonia could help to secure the health and wellbeing of future generations.

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New research on 21 patients with catatonia in the peripartum period finds that all episodes occurred following birth and 57 % of women had experienced obstetric complications.

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CRediT authorship contribution statement

AD, CAW and JPR designed the project. AD conducted the data extraction and drafted the manuscript. IJ and JG collected additional data following peer review. CAW, JPR, GS and RR revised the manuscript for important intellectual content.

Declaration of competing interest

JPR declares support from the Wellcome Trust. He has also received speaker fees from the Alberta Psychiatric Association and Infomed Research & Training Ltd. He has received book royalties from Taylor & Francis. He has received payment for reviewing from Johns Hopkins University Press.

The other authors declare no competing interests.

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