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Reevaluating the Place of Cycloid Psychoses: Case Study

Reavaliando o Lugar das Psicoses Ciclóides: A Propósito de um Caso Clínico

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RESUMO

Introdução: O conceito de psicose ciclóide foi descrito pela primeira vez por Karl Kleist. Mais tarde, Leonhard propôs a corrente conceptualização descrevendo três subtipos da doença e Perris desenvolveu os primeiros critérios diagnósticos operacionais. O diagnóstico de psicose ciclóide possui uma longa tradição na psiquiatria europeia, mas o conceito ciclóide não está explicitamente patente nos esquemas internacionais de diagnóstico (DSM 5 e ICD-10) suscitando um debate controverso quanto à sua utilidade e validade.

Objetivos: O presente artigo pretende, a partir de um caso clínico, abordar o conceito de psicose ciclóide enfatizando a sua importância à luz da psiquiatria atual, discutindo o uso do conceito e a sua validade clínica e preditiva.

Métodos: Os autores apresentam um caso clínico de psicose recorrente com total remissão interepisódica e afetação funcional mínima.

Resultados e Discussão: O artigo ilustra a importância de estarmos atentos ao diagnóstico de psicose ciclóide dado o seu prognóstico e tratamento distinto das restantes psicoses.

Conclusão: Enquanto esta perturbação, de incidência desconhecida, não for devidamente explorada, mais investigação será necessária dado o seu prognóstico favorável e a sua patofisiologia e tratamento potencialmente distintos.

Palavras-Chave: Psicose Ciclóide; Psicose Aguda; Reação Psicótica Breve; Perturbação Psicótica Transitória; Leonhard; Perris.

ABSTRACT

Introduction: *Karl Kleist primarily described the concept of cycloid psychosis. Leonhard set the basis for the current conceptualization of the disorder describing three overlapping subtypes and Perris developed the first operational diagnostic criteria. The diagnosis of cycloid psychosis has a long tradition in European psychiatry, but the cycloid concept is not explicitly included in standard international diagnostic schemes (DSM 5 and ICD-10) leading to a controversial debate about its utility and validity.*

Aims: *This article intends to evaluate, from a case study, the cycloid psychosis concept highlighting its importance and its clinical and predictive validity.*

Methods: *The authors present a case study of recurrent psychosis with total inter-episode*

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remission and minimal functional impairment.

Results and Discussion: *The article illustrates the importance of the diagnosis of cycloid psychosis given its distinct prognosis and treatment response compared with other psychoses.*

Conclusion: *While this disorder, of unknown incidence, is not well reported, it is worthy of further investigation and clinical attention given its generally favorable prognosis and potentially distinct pathophysiology and treatment.*

Key-Words: *Cycloid Psychosis; Acute Psychosis; Brief Psychotic Disorders; Transient Psychotic Disorders; Kleist; Leonhard; Perris.*

INTRODUCTION

"For me this is not a matter of some special diagnostic label, since it does not matter what names are used for these psychoses. However, as I understand it, the diagnosis of a mental illness includes a prognosis. When the diagnosis of cycloid psychosis is made, then it necessarily follows that a complete remission will occur and even if the illness recurs no defect will be left behind. This is, no doubt, very important." Karl Leonhard (1961)[1].

In the beginning of the twentieth century, when attentions were focused in the distinction of the idiopathic psychoses proposed by Emil Kraepelin, Karl Kleist proposed the concept of cycloid psychosis[2].

Kleist considered that some cases of atypical psychoses should be classified as independent nosological entities presenting with manic-depressive and schizophrenia-like features[3]. The concept of "cycloid marginal psychoses" was firstly presented characterizing atypical, acute, or recurrent psychoses. Then, two syndromes related to cycloid psychosis were introduced: *motility* and *confusional psychoses*.

Kleist considered that patients with cycloid psychoses had a predisposition to phasic illnesses with spontaneous remission that contrast with phases of confused excitement alternating with stupor, or hyperactivity alternating with inactivity. These illnesses, like manic-depressive insanity, presented with full recovery and no apparent deficits[2].

Cycloid psychoses had unique characteristics involving spells of excitement and inhibition, classically lasting two to four weeks contrasting with the extended phases of mania and melancholia in manic-depressive insanity[3].

According to Kleist, there were temperamental predispositions underlying cycloid psychoses mechanisms. He stated that both the cycloid and manic-depressive disorders seamed to rise from constitutional liability or dysregulation of affective, psychomotor, and cognitive functions, with comparable phasic courses and fluctuations between extremes of excitement and inhibition, with labile, polymorphous, and rapidly changing manifestations, typically without deterioration[3].

Karl Leonhard, Kleist's colleague, later introduced the concept of "endogenous atypical psychoses", as illnesses characterized by psychotic symptoms with an episodic pattern and without deterioration of function. He also investigated the "anxiety psychoses" described by Wernicke, specifically those presenting with

symptoms of perplexity, paranoid ideation, ideas of reference, and perceptual disturbances, with abrupt shifts in mood, going from anxiety to elation. He called these cycloid psychoses and proposed three major types: anxiety-elation, confusional excited-inhibited, and hyperkinetic-akinetic motility disorders[2].

All forms included interchange between a restricted-inhibited phase and an expansive-excited phase, involving a phasic and cyclic course and full inter-episode recovery[2].

In anxiety-elation psychosis only the affectivity is involved. The anxiety pole is associated with ideas of reference. The elation psychosis courses not only with expansive ideas, but, frequently, these patients want to make other people happy. They regularly believe that they are able to bring salvation to mankind by religious or political means. Anxiety psychosis is more common than elation psychosis[1].

The thinking process is affected in confusion psychosis. In the excited pole of the illness it shows as incoherence but very different from the incoherence of confused mania. Mutism is the distinguishing clinical picture in inhibited confusion psychosis. As these patients with inhibition of thought are not capable to understand the surrounding environment they become perplexed, so that "perplexed stupor" (Kleist) represents the typical syndrome of inhibited confusion psychosis. The inhibition of thought also leads to false interpretation[1].

The motility psychosis has two poles (hyperkinetic and akinetic) named according to the direction of the change in psychomotor activity. In both cases, the change implies an excess or deficiency of activity. There are no qualitative changes concerning the execution of movements. The increase or decrease of motor activity is related with the reactive and expressive movements that are based in psychomotor activity itself and do not need conscience consideration or intention. Both types of movement cease in akinesia. In complete akinesia the patient is almost motionless[1].

By the mid-twentieth century, Leonhard considered cycloid psychoses as distinct and as lying conceptually between episodic mood disorders and chronic schizophrenia[2].

Operational diagnostic criteria for cycloid psychoses were later developed by Carlo Perris and his colleagues[4]. The key features stated by Perris included confusion or di*stress*ed perplexity, and shifting, polymorphous symptoms. These investigators did not emphasize subtypes, as Leonhard, instead they followed a "symptom collection" approach, which is also found in ICD-9 and -10 and DSM-IV and DSM 5[2]. The diagnostic criteria for cycloid psychosis, according to Perris, can be found in Table I.

Several investigators following Leonhard and Perris have supported the nosological validity of cycloid psychosis. Others have considered it as a variant of major affective disorder closely related to bipolar disorder, as an atypical form of schizophrenia, or as excessively heterogeneous for reliable use. This lack of consensus has been associated with a decline in interest in cycloid psychosis since the 1980s, although the continuing lack of a satisfactory system for categorizing acute, remitting psychotic illnesses encourages renewed interest in the concept[2].

The international classification systems divide both clinicians and researchers. To increase the diagnostic validity and reliability patients

presenting with cycloid psychoses are categorized as acute polymorphic psychotic disorders, with or without symptoms of schizophrenia (F23 in ICD-10[5]) and brief psychotic disorder (298.8 in DSM 5[6,7,8]).

In this article, we report a case of cycloid psychosis and review the concept, nosological status, diagnostic features, associated clinical characteristics and the etiopathological variables involved in the condition.

Table I. Diagnostic criteria for cycloid psychosis by Carlo Perris[2,4,9].

1. An acute psychotic condition, not related to the administration or the abuse of any drug or to brain injury, occurring for the first time in patients between 15 and 50 years old;
2. The condition has a sudden onset with a rapid change from a state of health to a full--blown psychotic condition within a few hours or at most a few days;
3. At least four of the following:
   a. Confusion of some degree, mostly expressed as perplexity or puzzlement;
   b. Mood incongruent delusions of any kind, most often with a persecutory content;
   c. Hallucinatory experiences of any kind, often related to themes of death;
   d. An overwhelming, frightening experience of anxiety, not bound to particular situations or circumstances (pananxiety);
   e. Deep feelings of happiness or ecstasy, most often with a religious coloring;
   f. Motility disturbances of an akinetic or hyperkinetic type that are mostly expressional;
   g. A particular concern with death;
   h. Mood swings in the background that are not severe enough to justify a diagnosis of affective disorder;
4. No fixed symptomatological combination; on the contrary, the symptomatology may change frequently during the episode and have a bipolar characteristic.

CASE REPORT

The case we report is about a 45-year-old man that will be addressed as Mr. J.

Mr. J. was a construction worker who studied for only four years. He lived with his wife and teenage daughter. His family described him as a truly polite, respectful and hard-working man, though very shy, having some difficulties with interpersonal relationships.

Mr. J. had no psychiatric background up until March 2005, when he was admitted to our inward with a clinical picture of behavioural disturbance with sexual disinhibition and persecutory delusions. There was no history of substance misuse and he wasn't on medication when admitted to our department.

In March 2005, considering his financial problems, he went to work in The Netherlands. It was the first time he was living in another country and he had never travelled to anywhere so far away from home. After only one week abroad, he developed a clinical picture of behavioral disturbance with aggressiveness, sexual disinhibition, coprolalia and total insomnia. He was sent back to Portugal a day after the beginning of the symptoms and while waiting at the airport he started looking at the men around him with distrust stating they were trying to seduce him and intended to have sexual relationships with him. He also believed someone had mixed drugs in his cigarettes because they had a bad taste.

Already in Portugal, he was evaluated in the psychiatry emergency room and hospitalized in our inward.

On transfer to our unit he presented himself sleepy, dysarthric and mildly ataxic.

He was medicated with risperidone 3 mg (once a day) and observed by a neurologist in order to exclude any organic disease. Mr. J. was also submitted to a detailed investigation, including computerized tomography (CT) scan, hematologic and biochemical routine tests and a neuropsychological evaluation. Nothing was detected in CT and the analytic study showed no important changes. No substance misuse was detected. The MMSE (Mini Mental State Examination) and the clock drawing test didn't reveal any deficit (he punctuated 25/30* in the MMSE). The psychological evaluation diagnosed a mild intellectual disability.

Two days after his hospitalization, he showed an appropriate behavior, revealing no delusional activity. So he was discharged to outpatient follow-up being medicated with risperidone 3 mg (once a day). He abandoned this therapeutic regimen one week after he was discharged. He also abandoned the outpatient follow-up later.

In 2007, due to another financial crisis, Mr. J. decided to emigrate again, but this time went to work in Spain. A mere three days after he had arrived at the country, he developed a clinical picture of psychomotor restlessness, walking in the streets adrift stating he was following a light that was leading him somewhere. He expressed himself very anxious with the fact that he didn't understand the Spanish language. As a result, he was sent back to Portugal soon. Already at home, he maintained the behavior disturbance. He woke up his wife in the middle

* The MMSE scale adapted for the Portuguese population considers the existence of cognitive impairment when the patient punctuates 15 or less and is illiterate, 22 or less and has between 1 and 11 years of study, 27 or less if he had studied more than 11 years.

of the night stating he was extremely rich and that she should have sex with him immediately, if not, he would do it with their teenage daughter or even with their neighbour.
His wife accompanied him to the psychiatric emergency room and while waiting to be evaluated he presented himself restless and very disinhibited (he started masturbating in front of other men and spitting over everyone around him). He also assumed someone had mixed drugs in his food and that everyone in Spain was gossiping about him.
He was hospitalized for the second time and on transfer to our department he was perplexed, almost in mutism.
Again, he was submitted to a detailed investigation including CT scan, hematologic and biochemical routine tests and also electroencephalography (EEG). The clinical investigation didn't identify any relevant changes and there was no substance misuse detected.
He was medicated with risperidone 2 mg (once a day) with quick remission of the symptoms. On the 5th day of hospitalization, Mr. J. was discharged to our outpatient unit and medicated with risperidone 2 mg (once a day).

DISCUSSION

The diagnostic stability shown by cycloid psychoses seem to be reason enough to consider it as an independent nosological entity[9,10].
Leonhard's classification can help identify a subset of acute and transient psychotic disorders that have a distinctly favorable prognosis[8].
We believe this case is closest to the descriptions of cycloid psychosis, with symptoms predominantly from the Leonhard's excited–inhibited confusion subtype. In both episodes (2005 and 2007), Mr. J. clearly presented with two distinct clinical phases. Initially, he was excited, sexually disinhibited and talkative, with accelerated thought and incoherent speech, predominantly confused and hyperactive with fleeting ideas of reference. Then he fell into a perplexed, sleepy and dysarthric phase, being in mutism in the second episode.
The acute onset, the switching of polarity with polymorphous clinical symptoms, the short duration of the episodes – phasic course – with complete remission and no defect left behind support the diagnosis for this case[11,12].
This case also meets Perris's criteria for cycloid psychoses. Mr. J., a 45-year-old man, developed an acute psychosis of unknown cause with sudden change from health to psychosis within hours to a few days. He presented shifting polymorphic symptoms, with opposed polarities within a same episode, showing mood swings not sufficient to support a diagnosis of primary major affective disorder, deep feeling of happiness or ecstasy, confusion and paranoid features.
The case is difficult to categorize within the DSM5. Bipolar disorder with psychotic features would be improbable to present with rapidly fluctuating symptoms in multiple domains and multiple modes of perceptual disturbances without a mood disturbance preceding the episode. Schizoaffective disorder or schizophrenia would be rejected considering the complete remission of symptoms between episodes with no remaining defect. Other relevant categories of non-affective psychotic episodes in the DSM are brief psychotic disorder and schizophreniform disorder which

are based on time criteria and do not provide any psychopathologic distinctions on how to predict course. The ICD-10 category of "acute and transient psychotic disorders" specifies more detailed descriptive criteria although this can be criticized for its heterogeneity[13]. A cohort study over three years found a high level of diagnostic instability for this category[14]. The identification of cycloid psychoses as a subset of this category may be worthy of further study, with the hope of better characterizing this group and thereby improving prognostic validity[8].

Cycloid psychoses are only partially taken into consideration in the formal diagnostic systems and several studies that examined its nosological status have shown they do not correspond to any category included in modern classifications. Most cases of cycloid psychosis are diagnosed by these systems as brief psychotic disorder, schizophreniform disorder, schizoaffective disorder, mood disorder with psychotic features, or psychotic disorder not otherwise specified[15].

The diagnosis of a cycloid psychosis may be truly important essentially with respect to therapeutical consequences. Clinical experience has proved that the specific subform may play a considerable role in the pharmacotherapeutic approach. The acute therapy of anxiety psychoses usually requires neuroleptics, but the use of anxiolytic substances substantially controls the symptoms including the paranoid ideas. In most cases, the hyperkinetic expressive and reactive motions of the motility psychosis can be improved very fast by neuroleptics. The treatment of akinetic states is more difficult, as neuroleptics may not have benefits. They can be associated with severe akinetic complications, which are often mistaken for a malignant neuroleptic syndrome, thus the management of akinetic psychosis predominantly demands observation and the use of anxiolytic substances[16]. However, it should always be kept in mind that these phases also remit spontaneously. Electroconvulsive therapy (ECT) may be the first choice treatment if dangerous akinetic complications are presented. Neuroleptic long-term medication should not be prescribed to these patients although it frequently is, unfortunately, due to "schizophreniform" symptoms. Clinical experience indicates that for the phasic repetitions prophylactic treatment with lithium or anticonvulsive agents may be superior to neuroleptic maintenance therapy, although more studies should be carried out to support their benefits. In his initial studies, Perris found neuroleptics far less effective than lithium for prophylactic treatment of cycloid psychoses[17]. On the other hand there is evidence that after a sudden suspension of long-term neuroleptic treatment of cycloid psychoses the rate of relapse may increase[7,12].

Treatment of cycloid psychosis with antipsychotics, mood stabilizers, benzodiazepines, or electroconvulsive therapy has mainly an empirical basis. Antipsychotic drugs are the basis of therapy, and although atypical drugs appear to be especially valuable in patients with cycloid psychosis, more studies are necessary to prove their efficacy[15].

In the present case study, there was complete remission of the symptoms with low doses of risperidone, demonstrating the effectiveness of atypical antipsychotics in cycloid psychosis.

The patient abandoned the therapeutic after being discharged.
Additionally to Leonhard' studies[18], Beckmann *et al.* (1990)[19] have confirmed that the diagnosis of cycloid psychoses shows high stability with few shifts between the subforms[10,12].
The cycloid psychosis construct appears to have clinical validity and utility being easily differentiated from neighboring syndromes on psychopathological and outcome grounds. The clinical and heuristic value of the concept is irrefutable, as it defines a clinical syndrome with a relatively characteristic symptom pattern predicting good interepisode recovery. The interest of recognizing the concept lies on a correct differential diagnosis between patients with cycloid psychosis or with schizophrenia, something that could have dramatic therapeutic and prognostic implications. All these features reflect well the clinical and research importance of a concept that deserves to be included, on its own, in future formal diagnostic criteria of psychotic disorders[15].

CONCLUSIONS

The concept of cycloid psychosis refers to particular types of acute, phasic, polymorphic psychotic disorders. This concept may be a valuable alternative for some acute, good-prognosis psychotic disorders that are now very difficult to classify[2].
The cycloid psychosis concept should be reevaluated both for clinical diagnosis and to provide relatively homogenous disorders for genetic, physiological, neuroimaging, therapeutic, and other types of research[2].
Orlikov (2011)[20] throws out the question: should we reevaluate the place for cycloid psychosis in modern psychiatry?

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