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ESSAY

Catatonia: looking back and moving forward

Catatonia is a complex neuropsychiatric disorder with motor, affective and cognitive-behavioural manifestations. Understanding the relationships between these features has proven to be a challenge for psychiatrists and neurologists alike. Here we look back at the history of catatonia but also provide a modern perspective on how to understand the condition and what it can tell us both clinically and scientifically.

Catatonia can present with a bewildering constellation of symptoms. Some patients may be severely anxious, or unusually withdrawn to the point of being unwilling to speak, or they might display a fixed facial expression with no self-initiated movements. Some of them may repeat questions posed to them in a perseverative fashion and showed bizarre, exaggerated, and seemingly purposeless actions or movements. These motor phenomena (e.g. stupor, posturing, catalepsy, waxy flexibility, stereotypies, akinesia), affective signs (e.g. fear, aggression, anxiety, flat affect, affect incontinence, impulsivity), and cognitive-behavioural disturbances (e.g. mutism, autism, negativism, echolalia, echopraxia, grimacing, mannerism, rituals, automatic obedience) have all been observed in catatonia. Catatonia has been reported in 5–18% of patients in inpatient psychiatric units and 3.3% on neurology/neuropsychiatric tertiary care inpatient units.[1] Importantly, catatonia may be associated with potentially life-threatening circulatory collapse, respiratory collapse, renal failure, seizures, and coma. Timely recognition may therefore be lifesaving as catatonia tends to have a favourable prognosis once treated appropriately.

The term catatonia (from Greek *kata* = down + *tonos* = tension) was coined in 1874 by a German psychiatrist, Karl Ludwig Kahlbaum (1828–99). Its subsequent history is long and complicated but central to the beginning of clinical neurology and psychiatry, especially during the 19th century (see also Hirjak *et al.*[2]). At that time, there was not yet a separation of neurology and psychiatry. Indeed, motor disorders with strong psychological symptoms were formally characterized in the 19th century, including studies on Parkinson's disease (PD) (1817), multiple sclerosis (1868), Huntington's disease (HD) (1872), and Tourette syndrome (1844) by some of the most influential neurologists (e.g. James Parkinson, Samuel Alexander Kinnier Wilson, William Osler, George Huntington, Jean-Martin Charcot, and Gilles de la Tourette). Kahlbaum's original monograph *Catatonia or Tension Insanity*[3] published in 1874 falls into this conjoint neurological-psychiatric context. However, going beyond a predominant neurological description emphasizing primary motor system disorder with secondary psychological changes, Kahlbaum described a total of 21 psychotic patients presenting with flamboyant and partially bizarre signs including posture, mutism, negativism, and catalepsy. He particularly emphasized the strong affective component of catatonia driving the motor symptoms, observing that patients as a rule experienced turbulent emotional states prior to developing a catatonic episode characterized by stupor, catalepsy, waxy flexibility, stereotypies and echopraxia, i.e. prominent motor signs. According to Kahlbaum,[3(p30)]

> 'Mostly it is grief and worry and generally depressive moods and affections turned against oneself that give rise to catatonia. … Not infrequently, hypochondria and moods directed against the outside world, anger, sensitivity, irritability are also observed, and all the other melancholic symptoms, fear of poisoning, delusions of persecution, religious delusions of sin, etc. are often present as well.'

In 1877, Ewald Hecker from Görlitz (1843–1909), a German psychiatrist and student of Kahlbaum related a similar impression [Ewald Hecker (1877)]:

> 'This meant, however, that we would first have to expect a pronounced raving madness (which is only rarely absent in catatonia), and that furthermore the symptoms of melancholia attonita, flexibilitas cerea, mutism, refusal of food, confusion of names, stereotypy in speech and action, negativism, etc. would have to develop, at least in the greater majority, in the further course.'

Subsequently, this intimate relationship of catatonic motor signs with affective alterations as their driving factor was emphasized mostly in the Anglo-American tradition. In contrast, the continental European tradition stressed the relationship of catatonia to schizophrenia. More or less neglecting the affective component as driving force, this school of thinking reduced catatonia to purely motor signs (e.g. stupor, catalepsy, waxy flexibility, stereotypies and echopraxia) and emphasized that catatonia appeared to be almost exclusively associated with schizophrenia. Emil Kraepelin (1856–1926) and Eugen Bleuler (1857–1939) were the main proponents of categorizing catatonia with dementia praecox and later (from 1911) schizophrenia.[4] Kraepelin was concerned about the nosological classification of catatonia. In the sixth edition of his Textbook of Psychiatry catatonia became a subtype of dementia praecox. In Kraeplin's *Lectures on Clinical Psychiatry* from 1904 (p. 32) he wrote,

> 'Some thirty years ago a disease was described by Kahlbaum as catatonia, or 'insanity of rigidity', of which the most prominent symptom is a stiffness of the muscles, which would only be increased by outward interference. The disease should run through a series of different evolutions, and

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Figure 1 Catatonia patients by Eugen Bleuler (1857–1939).

> end at last in recovery or dementia. Overall, Kahlbaum's long-contested description has proven to be right, although I have to assume that the descriptions of disease summed up by him as catatonia are only special forms of dementia praecox.'

Overall, Kraepelin's work had an enormous impact on the understanding and on the concept of catatonia in the 19th and 20th century. Following Kraepelin and Bleuler (Fig. 1 shows Bleuler's patients with catatonia) catatonia was considered 'only' to delineate a subgroup within the spectrum of schizophrenia, i.e. schizophrenia with particularly 'prominent motor features' in the International Classification of Diseases (ICD) and DSM classification systems (Fig. 2). Therefore, the vast majority of psychiatrists narrowed Kahlbaum's original concept of catatonia as an independent disease entity with three distinct though closely intertwined aspects—affective, motor and cognitive-behavioural—down to a clinical syndrome with either prominent motor (continental European tradition) or affective (Anglo-American tradition) symptoms and signs.

This narrow understanding focusing on either motor or affective aspects of catatonia prevails in the current literature, an unfortunate fact which does not do justice to the original concept and potentially impeding appropriate clinical care. The overly strong emphasis on motor signs tends to lead to an underappreciation of affective and behavioural symptoms and signs, and may result in failing to recognize catatonia altogether, in particular if the motor component is not prominent, thereby delaying initiation of treatment (see below).

A certain neglect of the symptomatic complexity and even of catatonia as a separate mental disorder were even more accentuated by the introduction of antipsychotics in the 1950s. After the first administration of chlorpromazine by Jean Delay (1907–87) and Pierre Deniker (1917–98) in 1952 and the first description of antipsychotic-induced bucco-oral movements by Matthias Schoenecker in 1957 and tardive dyskinesia by Arild Faurbye (1907–83) and colleagues in 1964, catatonic motor and cognitive-behavioural symptoms and signs tended to be (mis)classified as side effects of antipsychotic medication, leading to a failure to appreciate that distinct motor signs may be a feature of the condition and not a side effect of its treatment.

However, the last 20–30 years have witnessed a resurgence of interest in the features and mechanisms underlying the syndrome. Catatonia was recognized to frequently co-occur with severe medical conditions, including delirium and autoantibody-related encephalitis.[1] Research also revealed that catatonia can occur independent of both schizophrenia and mood disorders such as major depressive or bipolar disorders.[2] This led to its recognition as an independent disease entity (at the same hierarchical level as schizophrenia or mood disorders) as it is now included in the ICD-11. The modern nosological reclassification circles back to the historical concept of catatonia as introduced by Kahlbaum. Catatonia is now recognized as complex amalgam of motor, affective, and cognitive-behavioural signs and symptoms—truly a psychomotor rather than purely motor phenomenon.

What exactly is meant by the term 'psychomotor'? In the authors' opinion, motor phenomena and psychic states/symptoms are intimately interconnected. For instance, very high levels of anxiety, beyond the control of the person experiencing it—as typically observed in catatonia—can lead to a complete motor 'freeze' with mutism, stupor and catalepsy. Primarily psychological (i.e. affective and/or cognitive changes) might thus rapidly bring about motor and cognitive-behavioural alterations, within minutes. As far as the interplay of motor, affective, and cognitive-behavioural signs and symptoms is concerned, there are intriguing similarities to neuropsychiatric disorders like PD or HD. In both these conditions, non-motor (affective and cognitive) symptoms may emerge before motor signs and influence each other in complex ways.

The distinction of motor versus psychomotor features is also mirrored in brain differences. PD, for instance, can be characterized by primary changes in the motor system such as the dopaminergic

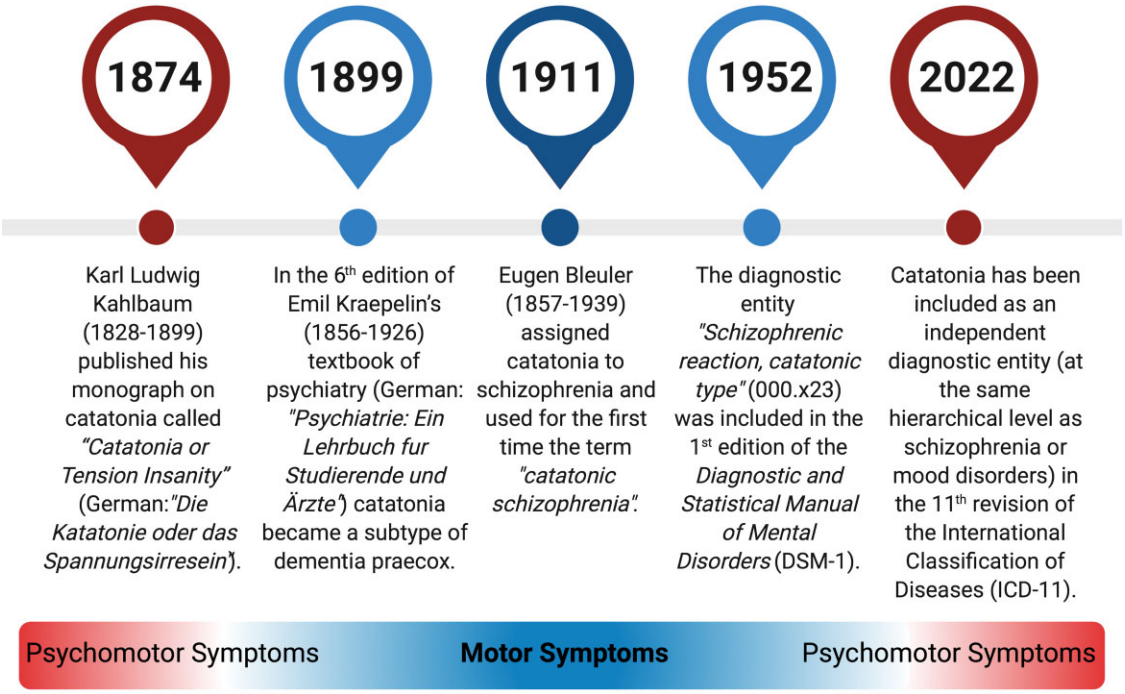

Figure 2 Important milestones in the history of catatonia.

substantia nigra and the frontostriatal motor loop involving the basal ganglia. But the pathology in PD may also extend to other higher-order cortical regions inducing affective and/or cognitive changes. Catatonia may also be associated with cortico-cortical alterations extending from orbitofrontal to motor cortex[2,5]: rather than primarily affecting substantia nigra-based dopamine, the syndrome may be related to raphe-nucleus based serotonin which, in turn, may down-modulate dopamine in the substantia nigra.[6] These more secondary motor subcortical-cortical changes may elicit the motor symptoms of catatonia that, like catalepsy and posturing, somewhat resemble but are not identical to akinesia and rigidity as observed in PD. This distinction between systems involved in motor vs psychomotor symptoms is also clinically relevant. A broader psychomotor view of catatonia provides a more comprehensive diagnostic spectrum by avoiding 'motor myopia', potentially enabling the clinician to start earlier treatment with, for example, benzodiazepines which can be effective in catatonia.

However, an understanding of catatonia as a psychomotor syndrome (with an inherent affective component) is currently not shared by all psychiatrists working on catatonia. Clinical rating scales for catatonia, with very few exceptions (e.g. Northoff Catatonia Rating Scale), emphasize motor signs and assess very few, if any, affective symptoms (mentioned only marginally in the widely used Bush Francis Catatonia Rating scale). It is therefore not surprising that most psychiatrists are familiar with *flexibilitas cerea* and posturing, but tend not to include affective (and also cognitive-behavioural) symptoms and signs in both clinical exploration and description of the clinical phenotype. Doing so would be important though given that patients often experience uncontrollable anxiety when for instance describing their catatonic motor symptoms in strongly affective terms.[7]

But do we really need an (additional) diagnosis ('catatonia') in patients presenting with severe affective symptoms? Aren't we dealing primarily with a mood disorder rather than catatonia? The distinction of catatonic motor signs from antipsychotic-induced motor abnormalities causes considerable discussion among psychiatrists and neurologists. Don't we need to treat severe affective alterations with antidepressants, which, in turn, will improve the catatonic symptoms? Or should we primarily treat catatonia? The same applies to motor signs: should we always initiate antipsychotic pharmacotherapy with the idea in mind to ameliorate schizophrenia as the root cause of the catatonic motor signs? Or do we risk further deteriorating a possible neuroleptic malignant syndrome or worsening antipsychotic-induced catatonia?[8]

These questions reflect the complexity of catatonia as a psychomotor disorder. We may want to return to the original concept of Kahlbaum but bring it up to date by augmenting it with a transdiagnostic and domain-based neuroscientific research agenda. There are no motor, affective or behavioural symptoms and signs in isolation, as we often implicitly assume. Instead, catatonia is a truly psychomotor disorder with multiple presentations highlighting the mutual interactions of psychological and motor alterations giving rise to a rapidly transforming clinical symptomatology. This was already appreciated by Kahlbaum in his monograph[3(p87)]:

> *'Catatonia is a cerebral disease with a cyclically changing course, in which the mental symptoms present in turn the picture of melancholy, mania, stupor (melancholia attonita), confusion and finally stupidity, of which overall mental pictures one or more may be absent, and in which, in addition to the mental symptoms, processes in the motor nervous system with the general character of convulsions (spasm) appear as essential signs.'*

Can we understand and translate Kahlbaum better to obtain more mechanistic insights in a time when we know so much more about the brain and its various neuronal circuits? Although modern views of brain function distinguish motor, affective, and cognitive networks/circuits, the symptomatic complexity of catatonia suggests that motor, affective, and behavioural symptoms and signs are not neatly separated but co-occur with multiple transitions. A spatiotemporal neuroscience[9] perspective might instead be useful. According to this view, the human brain is characterized by both an

intrinsic spatial topography and an intrinsic temporal dynamic that hold in spontaneous as well as stimulus-related activity. The temporal dynamic and spatial topography means that the brain constructs its own 'inner time and space' through and within its neural activity. Such a construction-based view shifts the focus from the different functions—cognitive, affective, and motor (including their neural correlates)—to their relationship based on the brain's own inner time and space,[9] its intrinsic topography and dynamic.

In catatonia, distinct symptom dimensions are primarily based on abnormal dwell times of specific states characterized by high within-network correlation of the sensorimotor, visual and default-mode networks.[10] Further, catatonia patients show increased static functional network connectivity in cerebellar networks along with abnormal low-frequency fluctuations over several other networks (e.g. basal ganglia, salience, default-mode, executive and visual networks).[10] Catatonia is therefore associated with distinct spatial and temporal dynamics of intrinsic neural network function. Since such dynamics operate across the boundaries of specific region or networks involved in particular functions, the syndrome manifests as a disorder of the affective-motor-cognitive relationship (rather than as disturbance of one single function like motor or affective or cognitive). This modern perspective, based on spatio-temporal neuroscience and its impact on psychopathology, may allow us to provide a more mechanistic understanding of Kahlbaum's original description of catatonia as a psychomotor disorder.

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Competing interests

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