

TOWARDS A VALID NOSOGRAPHY AND PSYCHOPATHOLOGY OF CATATONIA IN CHILDREN AND ADOLESCENTS

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Paraphrasing [Taylor and Fink \(2003\)](#), catatonia needs “a home of its own” in child and adolescent psychiatry. Limited but expanding literature supports that catatonia in children and adolescent can be identified reliably among other childhood conditions, is sufficiently common, treatable with the same specific treatments as adult catatonia (e.g., sedative drugs and electroconvulsive therapy), and can be worsened by other treatments (e.g., antipsychotics). Other findings in child and adolescent catatonia suggest that sex ratio and associated disorders may differ, and the proposed classification of [Taylor and Fink \(2003\)](#) needs modification. Adopting a broader diagnostic schedule may accommodate both child, adolescent, and adult catatonia. A *psychomotor automatism* variant should be included as a diagnosis, as well as specifiers for associated disorders such as acute nonpsychotic anxious state and pervasive developmental disorder. Duration of illness should be specified as acute or chronic. Regardless of associated psychiatric disorders, this chapter describes a new psychopathological model. Three main modalities of movement dysfunction in catatonic subjects are listed: (1) adherence to delusional ideas leading to a psychomotor automatism ([De Clérambault, 1927](#)); (2) resistance to delusional thinking or conviction; and finally (3) hyperanxious states. Case-vignettes illustrate the model, and future research directions are identified.

I. Introduction

Although Calmiel, in 1832, was the first to describe a sentinel report of malignant catatonia (Ainsworth, 1987), it was Kahlbaum (1874), who isolated the catatonic syndrome, characterized by the coexistence of psychic symptoms and motor symptoms resembling a muscular cramp. Kahlbaum (1874) stressed the frequent association of catatonia with depression or mania, as well as its link to organic conditions such as alcoholism, epilepsy, and syphilis. The catatonic syndrome became well known when Kraepelin and Bleuler, respectively, associated catatonia to dementia praecox and schizophrenia. Although both authors recognized that catatonic signs could appear in a variety of disorders, there was a subsequent tendency throughout the twentieth century to consider catatonia as a purely psychiatric condition associated with schizophrenia.

However, many reports published in the 1940s stressed that this view was too limited. For example, French psychiatrists Baruk (1959) and Ey (1950) pointed out that catatonia could also occur in organic conditions, manic episodes, melancholia, and hysteria; but their work remained unknown in the English medical literature. Similarly, research from the classic school of German psychiatry, particularly Kleist and Leonhard (Kleist, 1943), had little influence on international practice (Taylor and Fink, 2003). In the 1970s, the European position was endorsed by Morrison (1973), Abrams and Taylor (1976), and Gelenberg (1976), who seemed oblivious of the earlier French or German contributions. Finally, the broader view has been included in the Diagnostic and Statistical Manual, 4th ed. (DSM-IV), in which catatonia is still associated with schizophrenia, appears in a separate class as “catatonic disorder due to a general medical condition,” and is a specifier of affective disorders “with catatonic features” (APA, 1994). In the contemporary International of Classification Diseases, 10th revision (ICD-10), catatonia is only associated with schizophrenia and stupor with melancholia. A diagnosis of a medical condition is also possible under “organic catatonic disorder” (WHO, 1994).

Aims in this chapter are to formulate a specific nosography for catatonia in young people and present a supporting psychopathological model. First, the phenomenology of catatonia in children and adolescents will be reviewed.

II. Phenomenology of Catatonia in Young People

The estimated prevalence of catatonia in young people varies widely, that is, between 0.6% and 17% (Cohen *et al.*, 1999; Thakur *et al.*, 2003) but seems consistently lower than estimated rates of catatonia in adults, which range from

7.6% to 38% of admissions to psychiatric inpatient facilities (Taylor and Fink, 2003). Catatonia has been poorly investigated in child and adolescent psychiatry. In a previous review (Cohen *et al.*, 1999), only 42 cases were reported in the literature between 1977 and 1997. Takaoka and Takata (2003) listed 73 case-descriptions during the past 20 years (1982–2002). Most of the cases are adolescents. Systematic series over the last 5 years are limited to three: (1) Thakur *et al.* (2003) reported a consecutive series of 11 catatonic children and adolescents aged between 10 and 16 years in a pediatric clinic in Ranchi, India; (2) Wing and Shah (2000) reported 30 catatonic patients with a previous history of autism, in which catatonic features most often appeared between 10 and 19 years of age; and (3) Cohen *et al.* (2005) reported a prospective, consecutive series of 30 catatonic patients in two university clinics specialized in child and adolescent psychiatry in Paris, France. In an older study of childhood schizophrenia (Green *et al.*, 1992), catatonic symptoms were present in more than one-third (12 of 38) of cases. Future studies in larger samples are needed to determine the true prevalence of catatonia in psychiatrically disturbed children and adolescents.

In a case-series (Cohen *et al.*, 2005), more than half of the cases were of non-European origin (compared to approximately 20% of all inpatients in the study sites born to non-French native parents). A possible role of ethnic and/or cultural factors in the clinical expression of catatonia has been suggested in the adult literature (Leff, 1981; Varma, 1992). Given the high prevalence of catatonia (17%) in child and adolescent psychiatric inpatients in India, cultural factors may also play a role with child and adolescent inpatients (Thakur *et al.*, 2003). However, selection bias may explain some of the differences in cultural prevalence, and future researchers should consider this in their sampling. Cross-cultural comparative studies of childhood catatonia would be very informative in clarifying the role of cultural factors in the expression of catatonia in children and adolescents.

Phenomenology and associated diagnoses of catatonia are similar to those reported in the adult literature but relative frequency of associated disorder differed, with schizophrenia being the most frequent diagnosis (Cohen *et al.*, 1999). Moreover, the female-to-male ratio contrasts with that in adult studies. In young people, the majority of catatonic patients are boys (Cohen *et al.*, 1999; Takaoka and Takata, 2003), and catatonic schizophrenia appears to be a clinically relevant but understudied subgroup affecting males (Cohen *et al.*, 2005). In adults, women represent 75% of all cases of catatonia (Northoff *et al.*, 1999; Rosebush *et al.*, 1990). The difference in sex ratio might be a consequence of bias since affective disorders are overrepresented in samples of catatonic adults. Dhossche and Bouman (1997) compared frequencies of individual catatonic symptoms in pediatric cases (culled from the literature) versus adult cases. The estimated frequencies in adult catatonia were based on 463 catatonic cases pooled from seven studies. Symptoms in childhood versus adult catatonia were similar, except for incontinence that was not systematically reported in any of the

adult studies (see Table I). Incontinence is not a DSM-IV symptom of catatonia but frequently reported, at least in children and adolescents.

In Table II, symptom frequencies in a case-series of 30 catatonic adolescents are shown (Cohen *et al.*, 2005). Stupor and acute onset are symptoms present in most of the subjects with a mood disorder diagnosis, but they can be encountered in patients with schizophrenia as well; thus, they cannot be regarded as specific of a diagnostic

TABLE I
FREQUENCIES OF DSM-IV CATATONIC SYMPTOMS IN CHILDHOOD VERSUS ADULT CATATONIA

	Children % (N = 30)	Adults % (mean)	95% CI
Mutism	87	78	68–88
Posturing/grimacing	52	66	50–82
Stupor	80	66	45–87
Staring	49	57	35–79
Negativism	38	49	34–64
Rigidity	38	40	20–60
Stereotypy	24	37	22–52
Waxy flexibility	62	35	14–56
Echolalia/echopraxia	14	19	11–27
Excessive motor activity	14	15	10–20
Automatic obedience	10	10	4–16
Incontinence	45	–	–

Adapted from Dhossche and Bouman (1997).

TABLE II
BUSH–FRANCIS CATATONIA RATING SCALE MODIFIED FOR USE IN CHILD AND ADOLESCENT PRACTICE:
OCCURRENCE OF SYMPTOMS IN A SERIES OF 30 YOUNG PATIENTS WITH CATATONIA
(BUSH *ET AL.*, 1996; COHEN *ET AL.*, 2005)

Motor symptoms	(%)	Other symptoms	(%)
Catalepsy	67	Social withdrawal	90
Stupor	67	Mutism	80
Posturing	80	Mannerism	30
Waxy flexibility	60	Echophenomena	12
Staring	63	Incontinence	56
Negativism	80	Verbigeration ^b	33
Stereotypies	43	Refusal to eat	67
Psychomotor excitement	50	Social withdrawal	
Automatic compulsive movements ^a	53		
Muscular rigidity	67		

^a Including grimacing.

^b Meaningless and stereotyped repetition of words of note scoring is similar to that of Bush–Francis catatonia rating scale.

class. Automatic compulsive movements and stereotypies are highly suggestive (Cohen *et al.*, 2005; Kruger *et al.*, 2003) but not pathognomonic of schizophrenia.

A modified scale based on the Bush–Francis Catatonia Rating Scale (Bush *et al.*, 1996) was used. Six clinical items have been added based on Ey’s (1950) clinical studies on catatonia. Four of these (i.e., incontinence, refusal to eat, catalepsy, and automatic movements) were present in 56, 67, 67, and 53% of the subjects. Future research should focus on the validity and reliability of this modified scale in other child and adolescent samples.

III. Towards a Broader Nosography for Child, Adolescent, and Adult Catatonia

In a recent review, Taylor and Fink (2003) stated that catatonia should have “a home of its own” in psychiatric nosography, based on the following propositions: (1) it can be distinguished from other behavioral syndromes; (2) it is sufficiently common; (3) it improves with specific symptomatic treatments such as sedative drugs and electroconvulsive therapy (ECT); and (4) it can be worsened by other treatments (e.g., antipsychotics). Five amendments have been proposed to Taylor and Fink’s schedule in order to accommodate for findings in child and adolescent psychiatry and encompass a traditional French psychopathological entity (i.e., *psychomotor automatism*). Such a broadening may apply equally to youth and adult catatonia (Table III).

First, the addition of a fourth category of catatonia named psychomotor automatism was supported to help isolate the clinical presentation described later as the most frequent in adolescents (Cohen *et al.*, 2005). A typical case vignette is of a 16-year-old boy who had insidious onset of catatonic features, including automatic movements secondary to hallucinations—psychomotor automatism (De Clérambault, 1927), improves moderately when treated but continues to suffer a chronic course. This subtype is supported by a clinical study in adults. Kruger *et al.* (2003) performed a factor analysis of catatonic symptom distribution across four diagnostic groups; schizophrenia, pure mania, mixed mania, and major depression, and extracted a factor called “involuntary movements/mannerisms.” In this study, patients with catatonic schizophrenia exhibited more frequently symptoms represented in this factor. Second, catatonia secondary to general medical conditions, toxic state, or neurologic disorder should be grouped together as a specifier. When a catatonic syndrome occurs in the course of a general medical condition, it is associated with a cerebral impairment. This was the case for one patient who exhibited neuro-lupus and catatonia. The organic diagnosis focus of treatment may be very specific—plasma exchange and immunosuppressive medication in this case (Périsset *et al.*, 2003). Third, child and adolescent psychiatric literature suggests that catatonia can also be associated

TABLE III
PROPOSED CATEGORIES FOR DIAGNOSTIC CLASSIFICATION OF CATATONIA IN CHILDREN
AND ADOLESCENTS^a

Classification element	Category ^b
Catatonia	
DSM code xxx.1	Nonmalignant catatonia
DSM code xxx.2	Delirious catatonia (or excited catatonia)
DSM code xxx.3	Malignant catatonia
DSM code xxx.4	Psychomotor automatism (the main symptom is compulsive automatic movements)
Specifier for associated disorder	
DSM code xxx.x1	Secondary to a mood disorder
DSM code xxx.x2	Secondary to a medical condition (including toxic state and neurologic disorder)
DSM code xxx.x3	Secondary to a psychotic disorder
DSM code xxx.x4	Secondary to an acute nonpsychotic anxious state
Specifier for symptom course	Acute Chronic
Specifier for history of PDD	With a history of PDD

^aAdapted with permission from [Taylor and Fink \(2003\)](#).

^bIn bold, proposed modification in the field of child and adolescent practice.

with an acute nonpsychotic anxious state ([Thakur et al., 2003](#); [Ungvari et al., 1994](#)). Finally, two new specifiers were proposed to add: one was related to the course of the disease (acute versus chronic), and the second was related to a history of pervasive developmental disorder (PDD), due to its relative frequency in youth cases of catatonia ([Wing and Shah, 2000](#)). Although in the largest prospective series ([Cohen et al., 2005](#)), all patients ($N = 5$) with a history of PDD exhibited catatonic schizophrenia, the literature indicates that mood disorders can be associated with catatonia in patients with a history of PDD as well ([Ghaziuddin et al., 2005](#); [Révuelta et al., 1994](#); [Zaw and Bates, 1997](#)). Despite speculations of disease continuity between autism and catatonia ([Dhossche, 2004](#)), much more research is needed to elucidate the relation between autism and catatonia. This specifier is focusing research on this neglected symptom dimension in autism and to find better treatments for these conditions.

IV. Models of Catatonia

The phenomenology of the syndrome shows that catatonic symptoms can be classified into motor (e.g., posturing, catalepsy, waxy flexibility), behavioral (e.g., negativism, mutism, automatic compulsive movements), affective (e.g., involuntary

and uncontrollable emotional reactions, affective latence, flat affect, and withdrawal), and regressive symptoms (e.g., verbigeration, enuresis and encopresis, echophenomena). It is therefore extremely difficult to identify the subjective feelings experienced by a catatonic patient. For those who have an acute form of catatonia (e.g., some form of psychotic depression) (Cohen *et al.*, 1997; Northoff *et al.*, 1998), it is possible to ask patients retrospectively about such intrapsychic experience. For those who have a chronic form of catatonia (e.g., some forms of catatonic schizophrenia) (Cohen *et al.*, 1999), it is only possible with patients who do not show extreme mutism and negativism, when a careful interpersonal relation has been established.

Few models or hypotheses have been formulated, mostly biological, and all are somewhat unsatisfactory. Following the early experiments of bulbo-capnine-induced catatonia in animals with neocortex in 1928 (De Jong and Baruk, 1930), interests have focused on the role of (1) GABA_A receptor given the therapeutic efficacy of lorazepam, a GABA_A receptor potentiator (Bush *et al.*, 1996; Rosebush *et al.*, 1990); (2) *N*-methyl-D-aspartate (NMDA) receptor given that some catatonic patients, nonresponsive to lorazepam, were treated successfully with amantadine or memantine, both NMDA receptor antagonists (Northoff *et al.*, 1997; Thomas, 2005). Other neurochemical systems involved are the dopamine and the serotonine systems, although direct involvement remains controversial (Northoff, 2002a).

The most interesting attempt to model the pathophysiology of catatonia has been proposed by Northoff who has directed works on catatonia for years in adult patients, including clinical (e.g., Northoff *et al.*, 1999), pharmacological (e.g., Northoff *et al.*, 1997), electrophysiological (e.g., Northoff *et al.*, 2000a), and neuroimaging studies (e.g., Northoff *et al.*, 2000b). On the basis of comparing available literature on catatonia and Parkinson's disease (Northoff, 2002b) and catatonia and neuroleptic malignant syndrome (Northoff, 2002a), he proposed a functional neuro-anatomic model taking into account those similarities and differences regarding akinesia—a common feature in catatonia, Parkinson's disease and neuroleptic malignant syndrome—may reflect distinct modulation between cortico-cortical and cortico-subcortical relations. Northoff's model supports that clinical similarities between Parkinson's disease, neuroleptic malignant syndrome, and catatonia with respect to akinesia may be related to involvement of a dysregulation in a cortical-subcortical circuit (the so called “motor loop” between motor/premotor cortices and basal ganglia). Clinical differences in emotional and behavioral symptoms—that cannot be observed in either Parkinson's disease or neuroleptic malignant syndrome and define catatonia as a psychomotor syndrome—may be related with involvement of different cortical areas: orbitofrontal/parietal and premotor/motor cortices implying distinct kinds of modulation. This modulation is “vertical” in the case of the “motor loop” as opposed to “horizontal” in the case of psychic signs of catatonia. The affective, emotional, and behavioral symptoms in catatonia may be accounted for by dysfunction in orbitofrontal–prefrontal/parietal cortical

connectivity reflecting “horizontal modulation” of cortico-subcortical relation. Despite its deep and elegant proposals, Northoff’s model does not offer a more comprehensive psychological model of catatonia at the level of patient’s intrapsychic experience. Based on the clinical experience of catatonia in adolescents, a psychopathological model for catatonia was proposed and developed, which tends to explain the psychomotor symptomatology of the syndrome and to parallel the main clinical categories.

V. Psychopathological Model for Catatonia

There is a conspicuous lack of more comprehensive psychological models of catatonia at the level of patient’s intrapsychic experience. Findings in a few studies (Northoff *et al.*, 1998; Rosebush and Mazurek, 1999) as well as author’s own experiences give some insight into the subjective experience of catatonic patients. First, akinetic patients with catatonia appear unable to experience pain or fatigue despite prolonged posturing. This point is supported by possible complication of catatonia in skin injury lesions, even in young patients (Cohen *et al.*, 1999). Second, they appeared unaware of the objective position of their body or the consequences of their movements. Third, most of them reported intense and uncontrollable emotions, including one patient who had a blockade of his will with contradictory and ambivalent thoughts. Finally, all patients of Northoff *et al.* (1998) series remembered very well the persons who treated them on admission confirming that catatonic patients have no major deficit in memory and/or general awareness. This point was also highlighted by Rosebush and Mazurek (1999). The same experience has been shared with young patients except when a history of autism with no language does not permit retrospective psychological investigation. Similarly, except when catatonia is associated with a neurologic disorder, catatonic patients do not have abnormal neurological examination (Cohen *et al.*, 2005; Rosebush and Mazurek, 1999). Thus, neurological foundations are usually preserved in catatonic patients. Catatonic symptoms should be regarded as functional and also understood at the level of subjects’ experience resulting in catatonic motor dysfunction.

Based on the clinical experience of catatonia in adolescents, a psychopathological model for catatonia was proposed, which tends to explain the psychomotor symptoms of the syndrome, regardless of associated psychiatric disorders. First, a review of the main cognitive dimensions involved in voluntary human locomotion and movement is given as an aid in defining the specifics of catatonic psychopathology.

Figure 1 summarizes a cognitive model of voluntary human movements. Assuming an intact gross motor system, voluntary movement results from intentionality (or will), the behavioral planification, and the emotional context

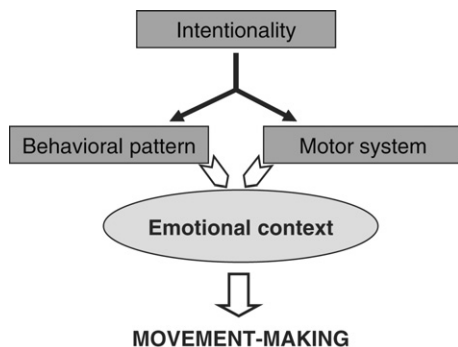


FIG. 1. A cognitive model of human voluntary movement (modified from Bloulac *et al.*, 2004).

(associated with the whole procedure), which in conjunction result in a voluntary movement. The intentionality belongs to the realm of human conscious activities, and its implication is not necessary for all type of movements (such as breathing). The behavioral planification per se is a complex procedure that involves, at minimum, attentional capacity, motivation skills, memory schemes and motor control, and computing (Bloulac *et al.*, 2004). A major but forgotten component (regarding catatonia) relates to the emotional context that is always present during a voluntary movement. The entire human repertoire can be encountered, such as sadness, anxiety, fear, anger, joy, and so on. Finally, the implication of the “motor system” is a truism but not under the scope of this chapter since there is evidence of normal neurological examination in catatonic subjects. However, its contribution to catatonic states may be suspected in cases of malignant catatonia secondary to extrapyramidal effects of neuroleptic prescriptions (Northoff, 2002a; Taylor and Fink, 2003).

Our psychopathological model that refers to the cognitive model of human locomotion described earlier, distinguishes three main modalities of subjective experiences that result in movement dysfunction in catatonic subjects: (1) adherence to delusional ideas that leads to a psychomotor automatism (De Clérambault, 1927), (2) resistance to delusional thinking or conviction, and (3) hyperanxious or hyperemotional states.

A. ADHERENCE TO DELUSIONAL IDEAS RESULTING IN CATATONIA

De Clérambault (1927) described the principle of psychomotor automatism. The most typical features of psychomotor automatism are automatic movements secondary to hallucinations and adherence to delusional ideas. The following case-vignette offers an illustration (Cohen *et al.*, 1999).

A was 16-year old when admitted to the inpatient unit for compulsive water drinking. At age 5, A started to be treated with psychotherapy for a nonautistic PDD. Although his intellectual skills were in the low average range, he remained in a regular school until age of 14 years. Then, he entered a special education program in which he received vocational training. One year later, he was sexually abused by a male peer. A began to isolate himself and started drinking a lot of water. A was first hospitalized for 2 months with symptoms described as compulsive behaviors: rituals to eat and drink and ordering rituals. He explained that he needed to drink a certain number of glasses of water in order to alleviate his anxiety. Fluoxetine (20 mg/day) and cyamemazine (50 mg/day) were started together with behavioral therapy for water control. As he was feeling better, he left the hospital for the summer holidays.

Three months later, A was readmitted to the unit in emergency because of life-threatening water intoxication. Few days before hospitalization, his parents said he was drinking up to 14 liters of water a day, shortly before admission. A was confused, complaining of headache and nausea, due to a cerebral edema confirmed by brain-imaging. Water restriction was sufficient to improve his confusion, but A exhibited catatonic signs, including stereotypies imitating the movement of drinking (even when water was not available), negativism, and catatonic excitement. He had numerous hallucinations including auditory orders to drink and to do specific movements resembling compulsive behaviors. He also had delusions with fears of HIV infection, or poisoning. A diagnosis of catatonic schizophrenia was made, and antipsychotics were administered. Chlorpromazine (250 mg/day) and then haloperidol (15 mg/day) failed to improve the patient's condition and instead induced numerous extrapyramidal effects. Thioridazine (350 mg/day) subsequently showed notable efficacy. A was able to attend behavioral therapy for water control and left the inpatient unit after 8 months. Follow-up was arranged at a day care hospital.

During a 2-year follow-up period, no catatonic symptom returned, while residual signs of schizophrenia, such as marked social withdrawal and lack of initiative, persisted. Pharmacotherapy was kept unchanged, but behavioral therapy was no longer necessary. A is now 22-year old and about to work in a supervised environment. Case 1 is typical of psychomotor automatism since he was totally adherent to his delusional voices and hallucinations during the acute phase resulting in automatic movements of drinking.

B. RESISTANCE TO DELUSIONAL THINKING OR CONVICTION RESULTING IN CATATONIA

Resistance to delusional thinking may also manifest as catatonia. In this scenario, subjects do not agree or follow voices and hallucinations giving orders, but desperately resist via bodily symptoms. These motor symptoms can be motor

rituals (very similar to automatic movements) designed to alleviate anxiety, or catalepsy and immobility designed to prevent any disaster. Case 2 is an exemplar (Cohen *et al.*, 1999).

C was a 13-year-old boy admitted to the unit for a paralytic catatonia. C had been treated since he was 4-year old for Asperger disorder. He had followed the regular curriculum of a supportive private school and attended psychotherapy twice a week until 11-year old when it was stopped as he was doing pretty well. A few months prior to admission, C developed severe compulsions and was diagnosed with obsessive-compulsive disorder. Clomipramine was started. His symptoms, however, were the beginning of his catatonic state. On admission, the catatonic syndrome was extreme due to a delay before hospitalization. C exhibited catalepsy with waxy flexibility and posturing lasting sometimes several hours. He also had muscular rigidity, negativism, stereotyped movements of the mouth (grimacing), hands, and shoulders, enuresis, and extreme acrocyanosis. C was not mute but exhibited blunted affect, disorganized thoughts, neologisms, and inappropriate smiles. He disclosed prominent delusions and hallucinations including voices of orders, tactile and olfactory hallucinations, and visions of skeletons. His compulsions appeared to be related to his delusions, and he was able to explain that he needed to stay immobile because he was terrified by voices ordering him to commit suicide.

Catatonic schizophrenia was diagnosed and antipsychotics were started after discontinuation of clomipramine. Catatonia did not respond to chlorpromazine (300 mg/day) or haloperidol (15 mg/day) and lorazepam (5 mg/day). A relay with thioridazine (200 mg/day) provoked a severe dystonic reaction that subsided after 1 week of medication discontinuation. After 4 months, ECT was proposed to the parents because of complications including skin injury lesions, but they refused the treatment. Amisulpride (800 mg/day) was associated with pack therapy¹ twice a week. This new therapeutic regimen led to moderate symptomatic improvement. After 6 months, C was able to leave the hospital and enter a day care unit.

Now 18-year-old C still exhibits residual symptoms both of the catatonic and schizophrenic spectrum. His treatment includes amisulpride (800 mg/day), biperidene (8 mg/day), psychotherapy twice a week, milieu therapy in a day hospital, and family therapy. Case 2 is typical since he desperately resisted to his delusional voices and hallucinations asking for suicide during the acute phase resulting in catalepsy, negativism, and immobility.

¹Pack therapy: Envelopment in damp sheets for 1 hour sessions with patient expressing cenesthetic sensations and spontaneous fantasies.

C. HYPERANXIOUS OR HYPEREMOTIONAL STATES RESULTING IN CATATONIA

This might be the most frequent subjective experience reported by stuporous and akinetic catatonic patients. Many authors have described hyperanxious experience in adults (Northoff *et al.*, 1998; Rosebush *et al.*, 1990) and adolescents (Thakur *et al.*, 2003; Ungvari *et al.*, 1994). This experience is usually associated with psychotic states, but psychosis is not required (Ungvari *et al.*, 1994). Case 3 is an exemplar of acute stuporous state associated with hyperemotionality and described how a brief neuroleptic medication induced a malignant form of catatonia (Cohen *et al.*, 1997).

M was a 15-year-old girl. One month prior to admission, M started to be sad, developed early-morning waking, and lost her appetite without any apparent cause. Severe psychomotor retardation worried her family. Her rapidly worsening mood symptoms prompted hospitalization. Delusional ideas concerning her own existence and a state of depressive stupor appeared a few days before her admission. When M was admitted to the inpatient psychiatric unit, she had lost 7 kg, showed severe psychomotor retardation, and exhibited catatonic posturing. Cyamemazine was started (75 mg/day). The next day, she was totally stuporous and refused food and water. Physical examination revealed a muscular rigidity, a trismus, a brief period of hyperthermia (38 °C), and an alteration of consciousness. Laboratory tests were all within normal limits, except creatin phospho-kinase (CPK) that had a concentration increased 12 times. Bacteriological samples, EEG, ECG, cerebrospinal fluid analysis, and head CT were normal. Within 48 hours after rehydration, neuroleptic retrieve, and Tropatepine² (20 mg/day), awareness was recovered, rigidity disappeared, and muscular enzymes decreased. She started speaking and moving spontaneously, though catatonic posturing and retardation remained. Delusion consisted of the absolute conviction that she was already dead, waiting to be buried, had no more teeth, had no more hair, and had a malformed uterus.³ The family was proposed ECT. Clinical improvement was notable after the first ECT in which she was then able to eat normally, speak fluently, and had no more delusive ideas. After the sixth treatment, she appeared to be lightly disinhibited and concerned with sexual matters. Although she had no other manic symptoms, it was decided to stop ECT. Rapid relapse led us to six more ECT sessions. Except headache following ECT, a few days of dysinhibition, and 3 weeks of mild confusion and disturbed memory, she showed no other secondary effects. Treatment at discharge associated amineptine (90 mg/day) and amisulpiride (600 mg/day).

² An anticholinergic drug available in Europe only.

³ In the French nosography, this delusional state has the name of Cotard's syndrome.

After the first ECT and later on, M's clinical status revealed that during the acute phase she was totally conscious but overcome by extreme anxiety as feeling she was already dead. It was wondered if her delusional ideas might have a meaning in her personal history. So far, she was asked to express the free associations that came in mind when they were evoked. Concerning the disappearance of her teeth, she felt surprised to think of her brother-in-law. She added that she would be ashamed to be given dental care by him and she started to cry every night since her sister's wedding and departure. On the idea of a reproductive malformation, she thought of guilty feelings associated with repeated masturbation that she had historically practiced from childhood until the beginning of her puberty. At 6 months follow-up, M had no more psychiatric symptoms. Memory impairments disappeared within 3 months, and she was able to go back to school 2 months after discharge. A manic episode occurred 1 year after the catatonic, warranting a mood stabilizer (carbamazepine 600 mg/day). She followed a psychodynamic therapy for 3 years and stayed 2 years in a residential setting for college student with psychiatric diagnosis. At 8 years follow-up, M continues to be euthymic, is planning to get married, and finishing her studies in social sciences.

Case 3 is typical of extreme emotional state resulting in catatonia. Anxiety was very high, and delusional ideas (e.g., being already dead) testify to the level of emotional involvement. Although comparing human behaviors to animal behaviors is an epistemological jump, whether these hyperanxious states resulting in catatonia may be related to the terror immobility reflex (e.g., freezing) described in the behavioral repertoire of many animals (Panksepp, 1998) is still not clear. According to Gallup and Maser (1977), comparing tonic immobility in animals and catatonic state in human may have an evolutionary significance as a survival mechanism. Catatonia may "represent fragments of primitive defences against predators that now misfire under conditions of exaggerated stress." They suggested that such conditions occur "in situations in which the person is frozen with fear." The fear can be external. But it is more frequently internal in human (as shown in case 3).

VI. Discussion

The proposed psychopathological model for catatonia is a modest attempt to give meaning and rationality to a psychomotor experience. It is based on patients' testimony of their subjective experiences during catatonic states when they accepted to share this experience during encounters. Figure 2 summarizes the proposal and attempts to link the main clinical presentation of the catatonic syndrome as exposed in the first part of this chapter.

Although this model may seem simplistic, it has clinical and heuristic validity. First, it fits well with the idea that the correlation of clinical form of catatonia and

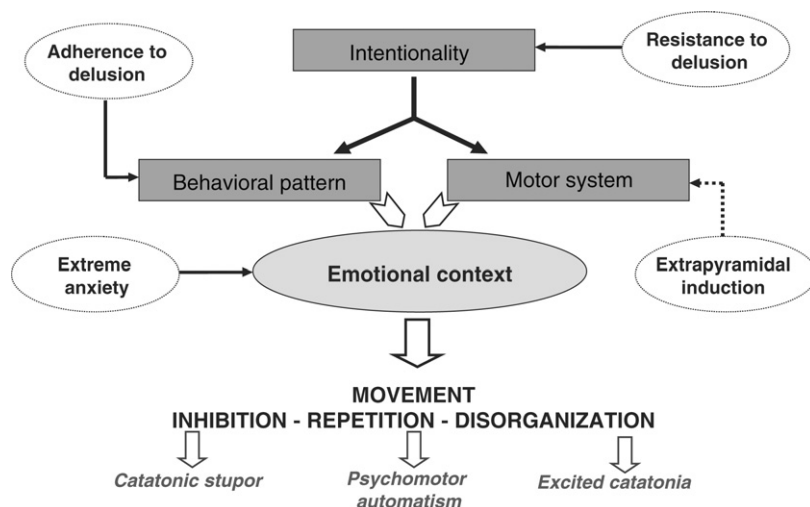


FIG. 2. A psychopathological model for catatonic symptoms. According to the model, catatonic signs are a consequence of: (1) a dysfunction of intentionality (e.g., when a patient resists to delusional ideas), (2) a dysfunction of the planification of behavioral patterns (e.g., when a patient strictly follows voices or hallucinations ordering movements—psychomotor automatism), and (3) a dysfunction of emotional regulation marked by extreme emotional involvement (e.g., in stuporous anxious states). Although direct correlation are not possible, these dysfunctions in movement making may explain the variety of catatonic motor signs (catatonic stupor, psychomotor automatism, excited catatonia).

associated psychiatric disorder is weak, although some direction may be highlighted: stupor and psychotic depression (Rosebush *et al.*, 1990; Taylor and Fink, 2003), automatic movements secondary to psychomotor automatism and schizophrenia (Cohen *et al.*, 2005; Kruger *et al.*, 2003), and excited catatonia and mania (Kruger *et al.*, 2003; Taylor and Fink, 2003). Second, the model allows for the diversity of clinical expressions and presentations of the catatonic syndrome. Third, the model gives a major role to subjective experience including emotional context compared to motor dysfunction. Finally, the model fits well with the fact that in many animals extreme fears can induce an immobilization reflex leading to a state resembling that of stupor.

However, the model has some major limitations. First, the model does not integrate data on functional anatomy of motor activities including regulation and planification. On the other hand, the neuronal basis of catatonia is not known and awaits further neuroscientific inquiry. To this respect, Northoff's proposals are interesting (described earlier). Second, a major dimension of human thinking, that is, temporality, has not been considered. This dimension is not included within the cognitive model of voluntary human movement proposed, although movement execution also includes a temporal paradigm (e.g., initiation,

execution, and then termination). Therefore, one can hypothesize that some catatonic patients also experience an alteration of temporality. There are some evidences showing that this dimension might play a key role in the subjective experience of catatonic patients. On one hand, in severe retardation, one can expect a suspension of temporality leading to stupor. In case 3, M had the conviction she was already dead, in some way “out of time.” Similarly, Northoff reported that many catatonic patients are not anymore aware of the consequence of their movements. Of course, this could be a consequence of an absence of awareness of their body position. It could also be interpreted as a lack of temporal awareness and anticipation. On the other hand, major increase of thinking speed may also result in a complete disorganization or a blockade of the sense of temporality. A 14-year-old girl with psychotic mania exhibited stupor and mutism. The diagnosis of mania was not possible at admission and was made when careful evaluation of functioning during the prior week was made with her mother. After recovery, thanks to a lithium trial, she explained that during the phase of stupor and mutism “thinking and ideas were that fast that I had no time to do anything even eating, talking, moving.”

Finally, the model does not encompass patients with no language, such as those with autism, since one does not have access to subjective experiences. In these subjects, it is to stress that the developmental course should be considered for diagnosis of comorbid catatonia, as suggested by [Wing and Shah \(2000\)](#) so that catatonia will not be overdiagnosed.

VII. Conclusions

Catatonia is an infrequent but severe condition in young people. While symptomatology and associated disorders are similar to those reported in the adult literature, findings differ in regard to the female-to-male ratio and the relative frequencies of associated disorders. There is a need for research in the field of catatonic schizophrenia in young people, as it appears to be a clinically relevant subgroup frequently resistant to therapeutic approaches. The inclusion of catatonia as a specific syndrome in the psychiatric nosography of both juvenile and adult practices may help its recognition and stimulate research in the field. Research should include psychopathological issues regardless the clinical or phenomenological presentation of the syndrome. In the psychopathological speculations that have not included the temporal dimension of movement and thinking, there are three distinguished modalities of movement dysfunction in catatonic subjects: (1) adherence to delusional ideas that leads to a psychomotor automatism ([De Clérambault, 1927](#)), (2) resistance to delusional thinking or conviction, and finally (3) hyperanxious states.

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