ECT FOR CATATONIA IN AUTISM SPECTRUM DISORDERS

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NACT NYKÖPING

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Disclosures

NONE

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International Review of Neurobiology, Volume 72

Annals of Clinical Psychiatry, Vol. 9, No. 4, 1997

Catatonia in an Adolescent with Prader–Willi Syndrome

Dirk M. Dhossche, M.D.,[1,3] and Nico H. Bouman, M.D.[2]

Catatonia in children and adolescents has received little research attention. Treatment and course of catatonia in an adolescent patient with Prader–Willi Syndrome are presented. Clinical features of a small series of published case reports of catatonia in children and adolescents are reported. The association between catatonia, Prader–Willi Syndrome, and other neurodevelopmental disorders is discussed.

KEY WORDS: Adolescents; catatonia; children; Prader–Willi Syndrome.

Table 1. Diagnosis, Age, and Sex in 30 Cases of Child and Adolescent Catatonia

Diagnosis	$N = 30$	Age (years) 7–12 $N = 7$	Age (years) 13–18 $N = 23$	Sex Male $N = 18$	Sex Female $N = 12$
Schizophrenia	3	0	3	2	1
Affective illness	6	1	5	3	3
Major depression	3	1	2	1	2
Bipolar illness	3	0	3	2	1
Organic illness	10	4	6	5	5
Medical illness[a]	7	3	4	2	5
Drug-induced condition[b]	3	1	2	3	0
Atypical psychosis[c]	11	2	9	8	3
With infantile autism	2	0	2	2	0
With mental retardation	1	0	1	1	0
Without developmental disorder	8	2	6	6	2

[a]Lyme disease ($N=1$); Wilson's disease ($N=1$); encephalitis lethargica ($N=1$); infectious mononucleosis ($N=1$); epilepsy ($N=2$); possible viral encephalitis ($N=1$).
[b]Steroids ($N=2$); haloperidol ($N=1$).
[c]This category includes cases with diagnoses of Atypical Psychosis Not Otherwise Specified and Brief Psychotic Disorder.

Table 3. Frequencies of Catatonic Signs in 30 Cases of Juvenile Catatonia as Compared to Adult Catatonics

DSM-IV catatonic signs	Children and adolescents % (N = 30)	Adults[a]	
		% (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
Urinary/faecal incontinence[b]	45	—	—

[a]Parameters based on a total number of 463 catatonic cases pooled from seven adult studies (16,38,48–52).

[b]Incontinence is not a DSM-IV catatonic sign.

Journal of Autism and Developmental Disorders, Vol. 28, No. 4, 1998

Brief Report: Catatonia in Autistic Disorders

Dirk Dhossche[1]

An adolescent case of catatonia superimposed on Autistic Disorder is presented. The literature regarding the association of the two disorders is briefly reviewed.

This case report adds to the few reports found in the literature on the association of autistic disorder and catatonia. Some psychomotor abnormalities in autistic disorders resemble "isolated" catatonic symptoms. The relationship with the catatonic syndrome is unclear. Both conditions have been associated with cerebellar and vermis atrophy (Courchesne, Yeung-Courchesne, Press, Hesselink, & Jernigan, 1988; Joseph, Anderson, & O'Leary, 1985; Wilcox, 1991). More empirical studies are needed to clarify clinical and biological aspects of the catatonic syndrome in autistic patients.

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Pediatric disorders associated with catatonia

Developmental Disorders

- Autism Spectrum Disorders (ASD)
- Childhood Disintegrative Disorder
- ID including Down Syndrome and Prader-Willi Syndrome
- Tic Disorders, Tourette Syndrome

Pediatric disorders associated with catatonia

Miscellaneous Conditions

- Anti-NMDARe (malignant catatonia + an antibody)
- PANDAS (catatonia + an antibody)
- Aseptic encephalitis (type EL) (autoimmune catatonia)
- Kleine Levin Syndrome (episodic adolescent catatonia)
- Psychogenic catalepsy (~ post-traumatic catatonia)
- Anaclitic depression (Spitz) (~ anaclitic catatonia)
- Pervasive Refusal Syndrome (exclusively European)
- Nodding Syndrome (exclusively in Uganda & South Sudan)

Prevalence of pediatric catatonia: studies since 1992

	N	Sample population	% cat
Green (1992)	38	Prosp, Child Schizo	32
Moise (1996)	13	Retrospective, ECT	46
Wing (2000)	506	Prospective, Autism	17
Thakur (2003)	198	Prospective, clinics	18
Billstedt (2005)	120	Prospective, Autism	12
Ohta (2006)	69	Prospective, Autism	12
Hutton (2008)	135	Prospective, Autism	5
Consoli (2009)	199	Meta-analysis, ECT	6
Ghaziuddin (2012)	101	Retro, At-risk inpts	18
Goetz (2014)	69	Retro, Ado 1st-Break	36

New-onset psychiatric disorders in individuals with autism

Vol 12(4) 373–390; 091650
1362-3613(200807)12:4

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ABSTRACT A follow-up study to at least the age of 21 years of 135 individuals with an autism spectrum disorder diagnosed in childhood and an IQ of over 30 was conducted. The study is distinctive in its large size, low attrition rate and use of systematic interviews to obtain clinical information. Questionnaires completed by caregivers asked about the development of new psychiatric disorders. For the 39 individuals with a possible new disorder, a detailed psychiatric assessment was undertaken through parental interview. Of all participants, 16 percent developed a definite new psychiatric disorder. A further 6 percent developed a possible new disorder. Five individuals developed an obsessive-compulsive disorder and/or catatonia; eight an affective disorder with marked obsessional features; three complex affective disorders; four more straightforward affective disorders; one a bipolar disorder; and one an acute anxiety state complicated by alcohol excess. There was no case of schizophrenia.

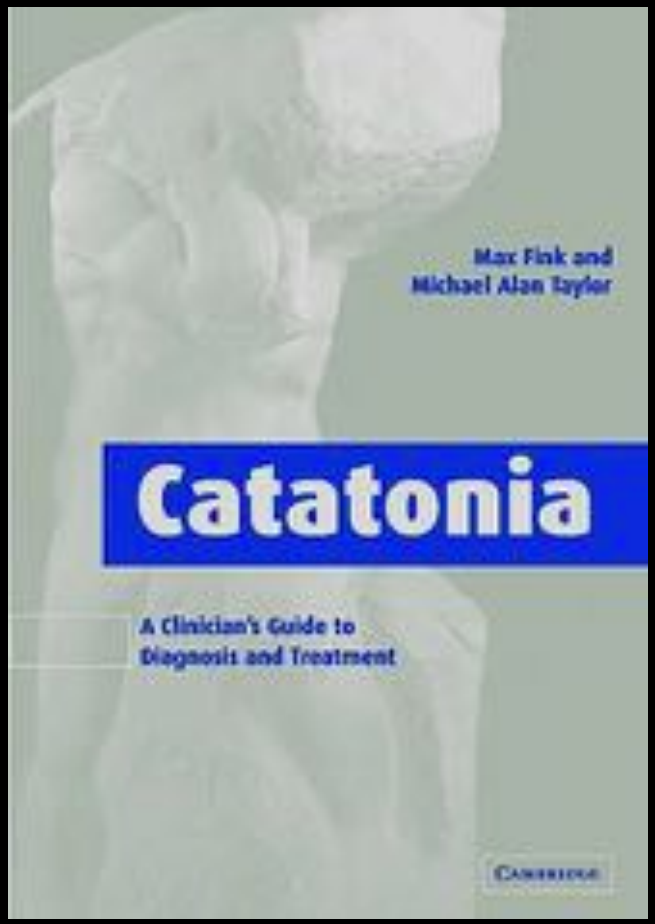
KEYWORDS
affective disorder; autism; catatonia; obsessive-compulsive disorder; schizophrenia

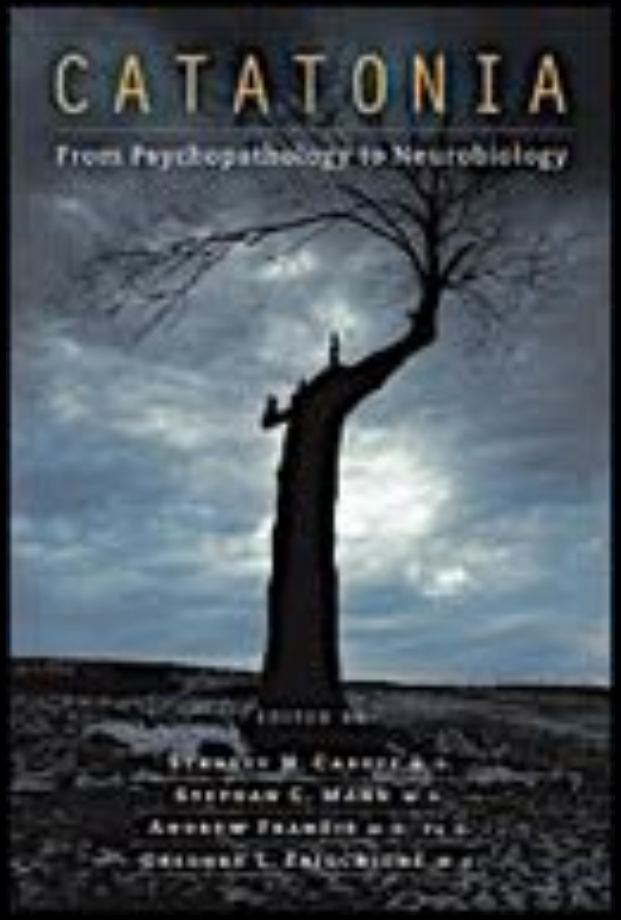
The presence of obsessive-compulsive behaviour and of catatonia (which mainly seemed to stem from obsessive-compulsive symptoms) may be particularly characteristic of individuals with autism and deserves special consideration both because of the dramatic nature of this presentation and because of the interventions sometimes used in catatonia. It has been suggested by some clinical investigators that catatonia might constitute an indication for electroconvulsive therapy (ECT: Dhossche and Stanfill, 2004; Ghaziuddin et al., 2005; Zaw et al., 1999). If the impression that catatonia develops as a result of obsessive-compulsive phenomena is correct, this would not seem a strong indication. It should be added that catatonia lacks a clear definition and, if it is to be diagnosed using a broad definition as Wing and Shah (2000) suggested, there would be a danger of moving too readily to heroic interventions of unproven value. Rather, the emergence of new obsessive-compulsive phenomena in adult life would seem to suggest the value of trying the pharmacological and/or psychological approaches that have been found to be effective in cases of obsessive-compulsive disorders unassociated with autism.

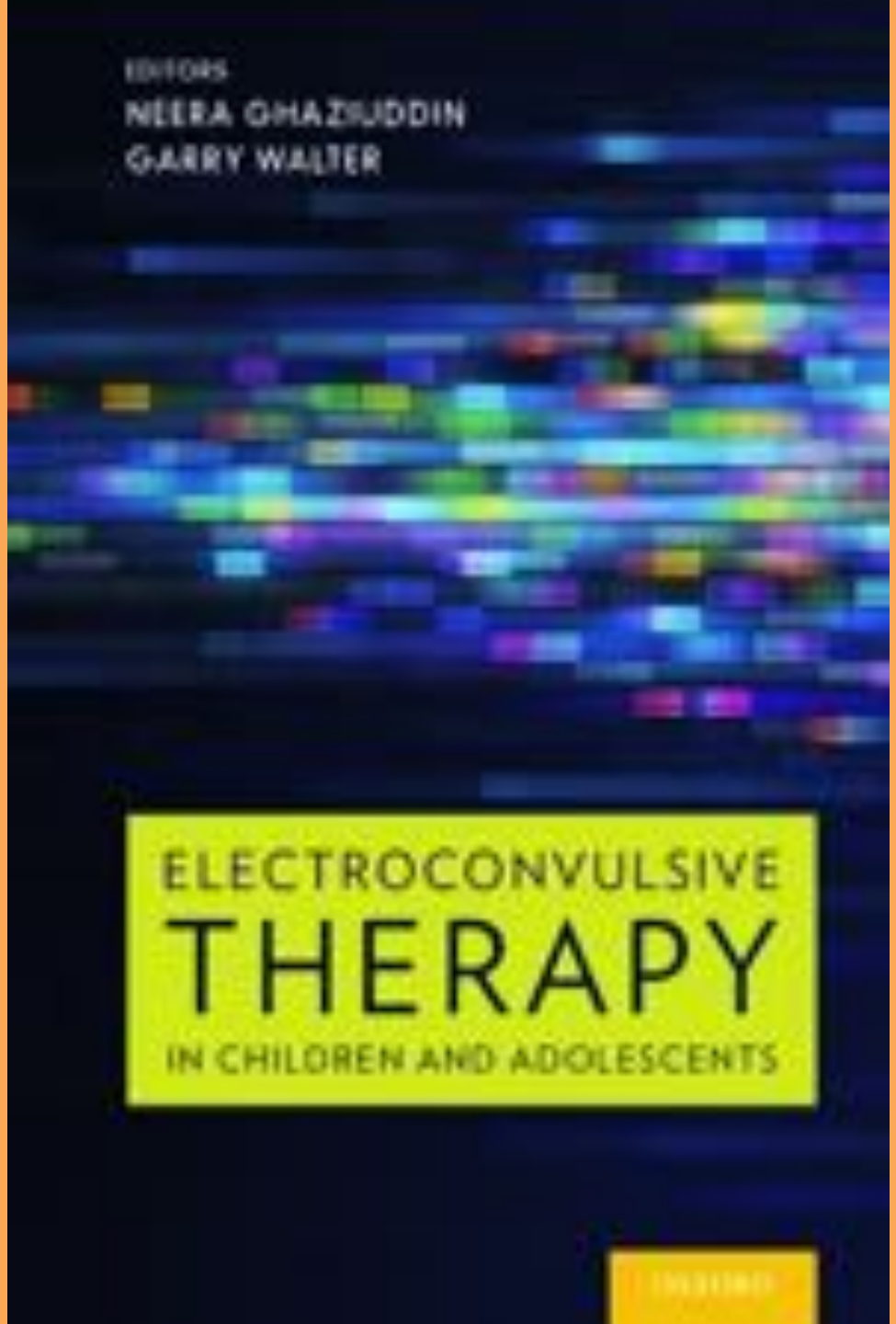
Mantra of the catatonia scholar

Catatonia is a poorly recognized syndrome (not a symptom!) & emergency that is eminently treatable with benzo's and ECT

Modern Catatonia Research






EDITORS
NEERA GHAZIUDDIN
GARRY WALTER
ELECTROCONVULSIVE
THERAPY
IN CHILDREN AND ADOLESCENTS

CHAPTER 11

ECT for Catatonia in Autism

DIRK M. DHOSSCHE, AND LEE E. WACHTEL

CHAPTER 12

ECT for Self-injurious Behavior

LEE E. WACHTEL AND DIRK M. DHOSSCHE

KEY POINTS

- Self-injurious behavior (SIB) poses a significant clinical challenge for many patients with autism or other intellectual disabilities.
- SIB may be maintained by operant or environmental factors; in addition, it may be associated with psychiatric, medical, and genetic disorders.
- Some forms of repetitive high-intensity, high-frequency SIB may represent a movement disturbance, best conceptualized as an alternate symptom of catatonia.
- This type of SIB may be exquisitely responsive to ECT with profound patient benefit.
- Multidisciplinary assessment of SIB should be completed before pursuit of ECT.
- Modifications to the ECT protocol may be necessary in pediatric patients with neurodevelopmental disabilities.
- Maintenance ECT is crucial, and poses unique challenges.
- Ethical and legal issues, lack of access to ECT, and stigma are salient obstacles to effective treatment.

AMY S. F. LUTZ

each day i like it better

AUTISM, ECT, AND THE TREATMENT OF OUR MOST IMPAIRED CHILDREN

FOREWORD BY DIRK DHOSSCHE, MD,
AND CHARLES KELLNER, MD

TREATMENT ISSUES

1 case with malignant catatonia

2 cases with catatonia and ASD

9-year-old boy with acute catatonia, one week after admission

J Autism Dev Disord
DOI 10.1007/s10803-014-2085-y

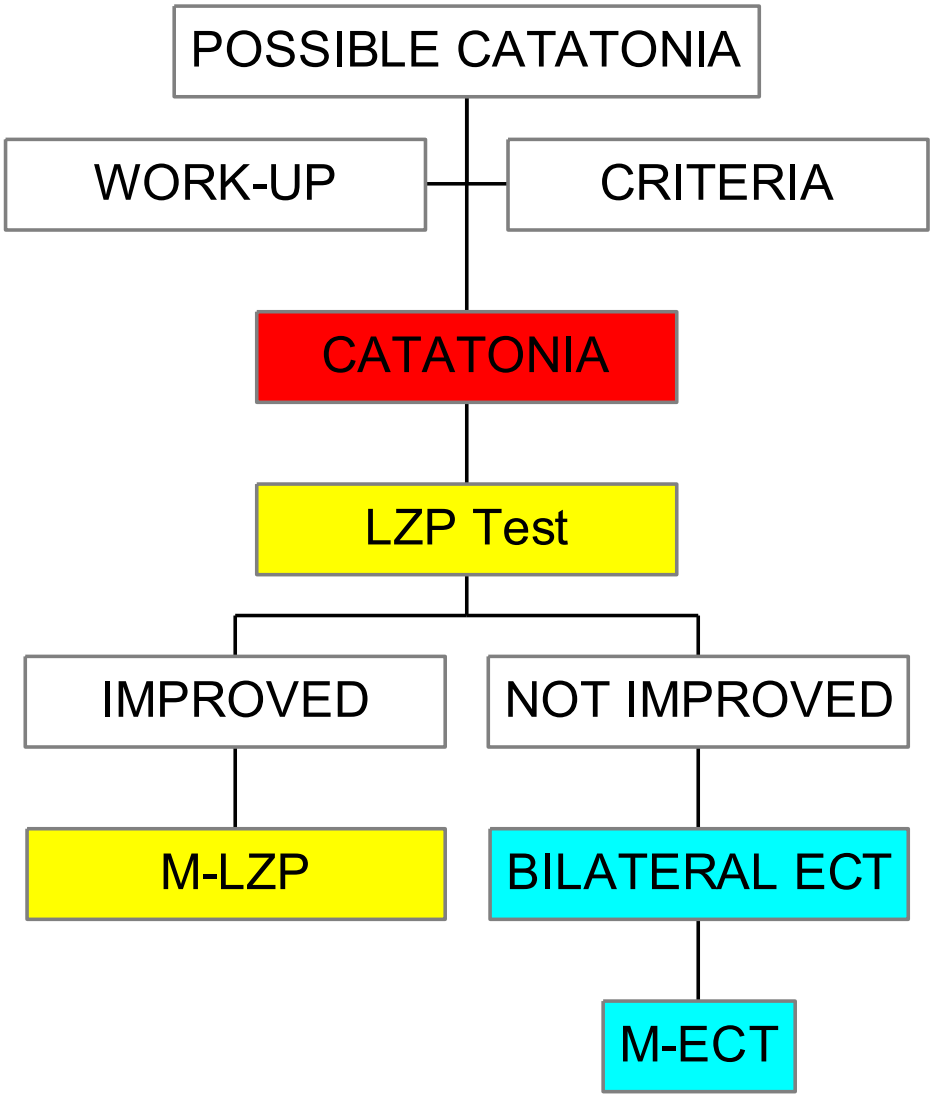
ORIGINAL PAPER

A Systematic Review of Interventions Used to Treat Catatonic Symptoms in People with Autistic Spectrum Disorders

Hannah DeJong · Penny Bunton · Dougal J. Hare

Abstract A systematic review was conducted to examine the efficacy of a range of treatments for autistic catatonia. The review identified 22 relevant papers, reporting a total of 28 cases including both adult and paediatric patients. Treatment methods included electroconvulsive therapy (ECT), medication, behavioural and sensory interventions. Quality assessment found the standard of the existing literature to be generally poor, with particular limitations in treatment description and outcome measurement. There is some limited evidence to support the use of ECT, high dose lorazepam and behavioural interventions for people with autistic catatonia. However, there is a need for controlled, high-quality trials. Reporting of side effects and adverse events should also be improved, in order to better evaluate the safety of these treatments.

EVALUATION, DIAGNOSIS, TREATMENT OF PEDIATRIC CATATONIA



10-year-old boy from California with high-functioning ASD

- **Acute onset OCD, 2 days after he became extremely upset after his best friend spit on his thermos during school, October 2012.**
- **He developed fears of being spit on, often of a delusional quality, and compulsions to protect himself, for the next 6 months.**
- **These problems required several admissions but worsened into catatonic episodes (mutism, refusal to eat/drink, negativism with incontinence) in March 2013.**

TREATMENTS DURING THE LAST YEAR

- Antibiotics, IVIG (x1) (PANDAS?)
- Olanzepine, quetiepine
- Citalopram, fluoxetine
- Olanzepine + fluoxetine
- LZP up to 16, then 24 mg (without sedation), but only with partial effect

a few years ago at his
birthday party

OPTIMAL TREATMENT?

MEDS: LZP + ARPZ

ECT

14-year-old boy from Texas with high-functioning ASD

- At term after a normal pregnancy.
- No speech until age 3
- He developed the tendency to look around out of the corner of his eyes and was not interested in friendships.
- Genetic testing was negative.
- Good participation in school, fluent conversation, performing ADL's independently

- **One year ago, at age 13, a sudden increase of abnormal movements including repeated turning of the head to the left, blinking, grimacing, stuttering, repetitive movements of the fingers, and rubbing of the eyes.**
- **Less speech and only in a high-pitched voice.**
- **Waxy flexibility when examined by a neurologist who found an otherwise neurologically intact adolescent (negative MRI brain, EEG, serum tests).**

- **A few weeks after onset of these symptoms, patient disclosed to an uncle that he was being bullied at school.**
- **Physical abuse and threats by peers were substantiated after an investigation by the school.**

- **Decreased food and fluid intake.**
- **Weight loss and poor sleep.**
- **Staring episodes, withdrawal, and episodes of compulsive hand washing and taking frequent showers.**
- **Preoccupation with death & crying spells.**
- **Tense and mask-like facial expression.**
- **Decreased writing skills and dropping grades.**

FAILED TREATMENTS

- SSRI's (fluoxetine, sertraline).
- Duloxetine, mirtazapine.
- Risperidone, aripiprazole, fluphenazine
- Clonidine, guanfacine.
- 4 mg of diazepam, 0.5 mg of lorazepam.
- A single dose of zolpidem (10 mg) caused agitation and increased tics.

14-year-old adolescent with high-functioning ASD and onset of catatonia since one year

J Autism Dev Disord
DOI 10.1007/s10803-014-2085-y

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Can trauma precipitate catatonia?

Yes

Past and current literature supports a role of traumatic events and overwhelming anxiety in the onset of catatonia

Acta Psychiatrica Scandinavica

Acta Psychiatr Scand 2012: 125: 25–32
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DOI: 10.1111/j.1600-0447.2011.01778.x

ACTA PSYCHIATRICA SCANDINAVICA

Clinical overview

The role of deprivation, abuse, and trauma in pediatric catatonia without a clear medical cause

Dhossche DM, Ross CA, Stoppelbein L. The role of deprivation, abuse, and trauma in pediatric catatonia without a clear medical cause.

Objective: Catatonia is considered a unique syndrome of motor signs, at times life-threatening when aggravated by autonomic dysfunction and fever, but eminently treatable with specific medical treatments, if recognized early. Catatonia commonly occurs in children and adolescents with a wide range of associated disorders. The role of deprivation, abuse, or trauma in the development of pediatric catatonia is examined.

D. M. Dhossche[1], C. A. Ross[2], L. Stoppelbein[3]

[1]Department of Psychiatry, University of Mississippi Medical Center, Jackson, MS, [2]The Colin A. Ross Institute for Psychological Trauma, Richardson, TX and [3]Department of Psychology, University of Alabama, Birmingham, AL, USA

- German psychiatrist Karl Kahlbaum, who coined the term catatonia in 1874, gave **trauma** a central role in catatonia in many young adult cases
- Kanner (1940) described children with **psychogenic catalepsy**
- **Anaclitic depression**, a condition found by Spitz (1944) in deprived institutionalized children, meets criteria for stuporous catatonia
- Leonhard (1960) considered **lack of communication with the mother or substitute mother** as an important risk factor for childhood catatonia

- Children, including those with autism, who experience **emotional and physical trauma** sometimes develop catatonia
- Swedish descriptions of contemporary traumatized **refugee children with a syndrome labeled Pervasive Refusal Syndrome** are those of children with classic catatonic syndromes

Advances in Psychiatric Treatment (2004), vol. 10, 213–219

Pervasive refusal syndrome

Bryan Lask

Abstract Pervasive refusal syndrome is a severe, pervasive and life-threatening disorder. Most commonly seen in girls between the ages of 8 and 15, although also affecting boys and younger age groups, it is characterised by a profound and pervasive refusal to eat, drink, talk, walk and engage in any form of self-care. A determined resistance to treatment is a striking component of the condition. The causes are unclear, but likely to be complex, multiple and associated with a sense of hopelessness. Treatment needs to be comprehensive and is based on supporting the child in recovering at her own pace, while ensuring physical safety and well-being. The prognosis is good, provided treatment is appropriate, but recovery tends to take a year or more.

Acta Paediatrica, 2005; 94: 1706–1707

ANNOTATION

Pervasive loss of function in asylum-seeking children in Sweden

GÖRAN BODEGÅRD

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Abstract

Presently, a couple of hundred children from traumatized asylum-seeking families in Sweden have developed severe loss of mental and physical functions without evidence of underlying disease. Of the 23 treated children treated at this clinic, 15 have recovered, three are improving and five are under initial care. Communication within the family is crucial from both pathogenic and salutogenic perspectives. A permanent residence permit, correcting the underlying situation of threat and insecurity, is a condition for good results from psychiatric treatment. In Sweden there is a lack of consensus and conflicting political and medical perspectives prevail regarding the "apathetic" children.

Conclusions: *Children living under unbearable life conditions can develop life-threatening depression-withdrawal stress reactions well known as pervasive refusal syndrome (PRS). This is also true of children in traumatized asylum-seeking families. Excellent results are achieved when the family's underlying fear and hopelessness can be erased and the treatment focuses on the traumatic experiences.*

- “The child is **totally passive, immobile, lacks tonus, withdrawn, mute, unable to eat and drink, incontinent, and not reacting to physical stimuli or pain**. Periods of panicky refusal and anxiety can precede or intervene with the stuporous state. Secondary symptoms may appear, such as **tachycardia, rise in temperature, edema, profuse sweating, reactivation of latent viral infection, skin ulcers and muscular atrophy**”
- No evidence of underlying medical diseases is found


WITH HER!
WHAT MAKES YOU
THINK THAT?

frontiers in Behavioral Neuroscience

HYPOTHESIS AND THEORY
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Resignation Syndrome: Catatonia? Culture-Bound?

Karl Sallin[1,2]*, *Hugo Lagercrantz*[2], *Kathinka Evers*[1], *Ingemar Engström*[3], *Anders Hjern*[4] *and Predrag Petrovic*[5]

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RS incidence in Sweden
2003-2005
2.8%
in 0-17 year old asylum seekers

- **Psychogenic catatonia is proposed to supply the best fit with the clinical presentation. Treatment response, altered brain metabolism or preserved awareness would support this hypothesis**
- **Epidemiological data suggests culture-bound beliefs and expectations to generate and direct symptom expression and**
- **It is argued that culture-bound psychogenesis can accommodate the endemic distribution**

Conclusions

- **Catatonia ALERT when there is a sharp increase of psychomotor symptoms in children, adolescents, & adults with autistic & developmental disorders.**
- **Catatonia can be precipitated by extreme stressors, life events, trauma, and abuse.**
- **ECT is an important treatment for catatonia in autism spectrum disorders (ASD).**