Medical Hypotheses xxx (2010) xxx–xxx

Contents lists available at ScienceDirect

Medical Hypotheses

journal homepage: www.elsevier.com/locate/mehy

The study of pediatric catatonia supports a home of its own for catatonia in DSM-5

Dirk Dhossche [a,*], David Cohen [b], Neera Ghaziuddin [c], Charmaine Wilson [a], Lee Elizabeth Wachtel [d]

[a] Department of Psychiatry, University of Mississippi Medical Center, 2500 North State Street, Jackson, MS 39216, USA
[b] Université Pierre et Marie Curie, Service de Psychiatrie de l'Enfant et de l'Adolescent, Groupe Hospitalier Pitie-Salpetriere, 47-83 Boulevard de l'Hôpital, 75651 Paris Cedex 13, France
[c] Department of Psychiatry, University of Michigan, Rachel Upjohn Building, 4250 Plymouth Road, Ann Arbor, MI 48109-5734, USA
[d] Kennedy Krieger Institute/Johns Hopkins School of Medicine, 707 North Broadway Street, Rm. 232, Baltimore, MD 21209, USA

ARTICLE INFO

Article history:
Received 15 April 2010
Accepted 17 July 2010
Available online xxxx

SUMMARY

The study of pediatric catatonia has not received much attention. During the last few years, progress has been made in delineating this syndrome in children and adolescents across a wide range of disorders. Catatonia is a potentially life-threatening but treatable syndrome that also occurs in children and adolescents with autistic, developmental, and tic disorders, and in its idiopathic form. In many of these cases, catatonia cannot be accounted for by an associated psychotic, affective, or medical disorder. These findings are imminently relevant for classification where catatonia is currently restricted to sections of the psychotic, affective, or medical disorders. Catatonia should always be the primary diagnosis in children, adolescents, and adults, as specific treatments for catatonia, i.e., benzodiazepines and electroconvulsive therapy, lower risk of worsening catatonia or precipitating Neuroleptic Malignant Syndrome when antipsychotic medications are used as first-line or sole treatment. The creation of a separate diagnostic class for catatonia is the safest approach to ensure proper diagnosis and treatment of this syndrome in patients of all ages and the best approach to promote research.

Introduction

Catatonia is a psychomotor syndrome, characterized by motoric immobility sometimes alternating with excessive motor activity (that is apparently purposeless and not influenced by external stimuli), extreme negativism or mutism, peculiarities of voluntary movement, or echolalia or echopraxia. Catatonia was originally described in adults by Kahlbaum in 1874 as a unique syndrome [1]. Kraepelin incorporated catatonia as a type of schizophrenia [2] although subsequent studies have shown that the syndrome occurs in adult patients with psychotic, affective, drug-induced, and medical disorders [3,4]. Unfortunately, catatonia remains erroneously perceived as uniquely associated with schizophrenia [5]. Benzodiazepines and electroconvulsive therapy (ECT) relieve catatonia in most instances. Associated conditions may require different and additional treatments.

One of the most important changes for clinical practice in the future DSM-5 concerns the further separation of catatonia from schizophrenia [6]. Catatonia is currently found in several categories of DSM-4 [7], and this may add to diagnostic confusion and poor selection of effective treatment for catatonia. First, catatonia due to a general medical condition (code 293.89) is listed as a distinct entity in the section on medical conditions. Second, the catatonia subtype of schizophrenia (code 295.20) is one of the original Kraepelinian forms of schizophrenia listed since the first DSM edition in 1952. Third, catatonia is an episode specifier for depressive disorder and bipolar disorder but without separate diagnostic codes. Fourth, the diagnosis of NMS, which some consider a form of malignant catatonia [8,9], is listed separately as a medication-induced movement disorder.

The degree of independence that catatonia should be allotted in DSM-5 is actively debated. Some [5,10] believe that catatonia deserves a home of its own in psychiatric classification, as originally proposed by Taylor and Fink [11] and that catatonia's divorce from schizophrenia and its recognition as an independent syndrome, akin to delirium, are needed in the next psychiatric classification. Independence of catatonia would increase timely recognition of its frequent comorbidity with affective disorders and general medical disorders, and increase attention to specific diagnostic and therapeutic measures (specifically high-dose benzodiazepines and ECT) that are commonly overlooked. Catatonia research would also benefit from greater convergence with the existing body of research on repetitive behavior disorders.

Others [12] warn against completely severing catatonia from the major psychotic and mood disorders as this may lead, unintentionally, to continued neglect of catatonia as a valid syndrome, to continued exclusion from vital research on the significance of psychomotor phenomena as a symptom dimension of psychotic and mood disorders, and to classification problems in schizophrenia given their historical associations.

* Corresponding author. Tel.: +1 601 984 5805; fax: +1 601 984 6965.
E-mail address: dr6340451@pol.net (D. Dhossche).

0306-9877/$ - see front matter © 2010 Elsevier Ltd. All rights reserved.
doi:10.1016/j.mehy.2010.07.029

The recommendations of the DSM-5 Psychosis Work Group have recently been posted on-line (www.dsm5.org, accessed on 14 April 2010). The Work Group aims to improve the recognition, treatment, and study of catatonia. The creation of a new diagnostic class for catatonia with specifiers for comorbid conditions versus the use of specifiers in the psychotic, affective, and medical disorders are offered as options. The Work Group finds catatonia primarily linked to schizophrenia and antipsychotic medications not contraindicated in catatonia. The creation of a new diagnostic class in the psychoses chapter for catatonia is not supported. Instead, the use of catatonia specifiers in the psychotic, affective, and medical disorders is favored, thereby eliminating the traditional catatonic subtype of schizophrenia (295.2) and the freestanding catatonic disorder due to a general medical condition not elsewhere classified (293.89), leaving catatonia uncoded.

Pediatric catatonia

The recommendations of the Work Group are not consistent with findings in pediatric catatonia [13,14]. Pediatric studies [15–22] listed in Table 1 and two case-vignettes [23,24] show catatonia to be a treatable syndrome that is hidden in plain sight across a wide range of disorders. Adolescents with schizophrenia and affective disorders have a 60-fold increased risk of premature death, including suicide, when compared to the general population of same sex and age [25]. Various medical-neurological disorders and substance-induced conditions are associated with pediatric catatonia requiring different and additional treatments [26–28].

Catatonia also occurs in children and adolescents with autistic [15,20,22,24,29–44], developmental [45–48], and tic disorders [49,50], and in its idiopathic form [23,51–56]. In many of these cases, catatonia cannot be accounted for by an associated psychotic, affective, or medical disorder. There are risks in severe morbidity and mortality of acute catatonia when not promptly recognized and treated as catatonia may worsen or Neuroleptic Malignant Syndrome may ensue when prescribing antipsychotic medications as first-line or sole treatment [57–62].

Case of adolescent malignant catatonia [23]

A 16-year-old adolescent with normal development and unremarkable psychiatric history presented in the emergency room with one month onset of bizarre behavior, motor abnormalities, and hallucinations after recovering from pneumonia. A medical work-up for suspected encephalitis was negative. CPK levels were low. Psychiatric examination was positive for muteness with some echolalia, echopraxia, facial grimacing, psychomotor agitation alternating with psychomotor retardation, and episodes of facial flushing and profuse sweating. The patient was diagnosed with idiopathic malignant catatonia. Administration of 1–2 mg of lorazepam did not bring relief and ECT was pursued. ECT, administered on three consecutive days, decreased agitation and unresponsiveness. On the fourth day, the patient showed a dramatic improvement with resolution of most catatonic symptoms and near-return to baseline function. He was discharged from the hospital after six ECT treatments and received one outpatient ECT treatment. He has remained free of psychiatric symptoms at two-year follow-up.

Table 1
Prevalence of pediatric catatonia: review of the literature since 1992.

Authors (year)	Sample size	Design sample population	Percentage with catatonia
Green et al. (1992) [18]	38	Prospective Childhood schizophrenia	32
Moise and Petrides (1996) [19]	13	Retrospective ECT	46
Wing and Shah (2000) [22]	506	Prospective PDD	17
Thakur et al. (2003) [21]	198	Prospective Psychiatric clinics	18
Cohen et al. (2005) [16]	4976	Prospective Psychiatric clinics	0.6
Billstedt et al. (2005) [15]	120	Prospective PDD	12
Ohta et al. (2006) [20]	69	Prospective PDD	12
Consoli et al. (2009) [17]	199	Literature review ECT	6

Case of adolescent malignant catatonia in autism [24]

A 15-year-old male with high-functioning autism developed gradual onset of slowness, loss of verbal skills and delayed task completion followed by development of rigidity, posturing, waxy flexibility and negativism in the forms of food refusal and urinary retention. Psychiatric assessments revealed no evidence of affective or psychotic pathology, and the boy's symptoms were attributed to autism and mental retardation after thorough evaluations. A diagnosis of catatonia was finally made after 10 months of frank catatonic symptoms, at which point the patient required constant cardio-respiratory monitoring because of severe autonomic instability. His parents' insurance carrier initially denied coverage for ECT stating that he had neither a psychotic nor affective illness. ECT was nonetheless pursued, and all catatonic symptoms remitted. Exhaustive subsequent psychiatric evaluations failed to reveal any hint of affective or psychotic pathology. This patient presented with malignant catatonia alone, and lack of rapid diagnosis and treatment nearly cost him his life.

Implications

A new diagnostic class for catatonia, such as catatonia not otherwise specified (NOS), is warranted in order for catatonia to be diagnosable in children and adolescents without a comorbid diagnosis of schizophrenia, affective disorder, or medical disorder. Such a classification would also prevent premature closure on evolving research on pediatric catatonia in autistic, neurodevelopmental, and tic disorders, and of idiopathic catatonia.

Where should catatonia NOS be placed? The safest solution should increase the visibility of acute catatonia as a life-threatening emergency in all patient populations – adult and pediatric – and lessen the risk of worsening catatonia or precipitating Neuroleptic Malignant Syndrome by favoring traditional anti-catatonic treatments, i.e., benzodiazepines or ECT, over antipsychotic medications as first-line or sole treatment [57–62]. We believe the best solution to be the creation of a separate catatonia class (*with own code*) with specifiers for associated psychotic, affective, medical, and toxic conditions; a home of its own for catatonia, as originally proposed by Taylor and Fink [11]. Taking catatonia out of schizophrenia and psychosis may pose problems for the classification of schizophrenia given their traditional associations [2,12] but this should be balanced by the benefits of increased safety and decreased complications. This would best serve catatonia research as there is no indication that a significant number of schizophrenia or psychosis researchers are wedded to studying catatonia [63].

The proposal of Taylor and Fink seeking independent status of catatonia should be amended to include a catatonia NOS specifier allowing proper diagnosis of pediatric catatonia in various disorders (especially the autistic and neurodevelopmental disorders)

Please cite this article in press as: Dhossche D et al. The study of pediatric catatonia supports a home of its own for catatonia in DSM-5. Med Hypotheses (2010), doi:10.1016/j.mehy.2010.07.029

outside the major psychotic, affective, and medical disorders. The creation of a new catatonia class with specifiers for associated conditions as well as for catatonia NOS seems the safest and best approach. Mention should be made that there is a severe form of catatonia, i.e., malignant catatonia, characterized by fever and autonomic disturbances, that the onset can be acute or gradual, and the course chronic or episodic [3,4].

Conflict of interest statement

None declared.

References

[1] Kahlbaum K. Die Katatonie oder das Spannungsirresein. Berlin: Verlag August Hirshwald; 1874.
[2] Kraepelin E. Psychiatrie: Ein Lehrbuch fur Studirende und Aerzte. 5th ed. Leipzig: Barth; 1896.
[3] Caroff S, Mann S, Francis A, Fricchione G. Catatonia. From psychopathology to neurobiology. Washington, DC: American Psychiatric Publishing, Inc.; 2004.
[4] Fink M, Taylor M. Catatonia. A clinician's guide to diagnosis and treatment. Cambridge: University Press; 2003.
[5] Fink M, Shorter E, Taylor M. Catatonia is not schizophrenia: Kraepelin's error and the need to recognize catatonia as an independent syndrome in medical nomenclature. Schizophr Bull 2010;36:314–20.
[6] Heckers S, Tandon R, Bustillo J. Editorial: catatonia in the DSM—shall we move or not? Schizophr Bull 2010;36:205–7.
[7] American Psychiatric Association. Diagnostic and statistical manual of mental disorders. 4th ed. Washington, DC: American Psychiatric Association; 1994.
[8] White D. Catatonia and the neuroleptic malignant syndrome – a single entity? Br J Psychiatry 1992;161:558–60.
[9] Fink M, Taylor M. Neuroleptic malignant syndrome is malignant catatonia, warranting treatments efficacious for catatonia. Progr Neuropsychopharmacol Biol Psychiatry 2006;30:1182–3.
[10] Rosebush P, Mazurek M. Catatonia and its treatment. Schizophr Bull 2010;36:239–42.
[11] Taylor M, Fink M. Catatonia in psychiatric classification: a home of its own. Am J Psychiatry 2003;160:1–9.
[12] Ungvari G, Caroff S, Gerevich J. The catatonia conundrum: evidence of psychomotor phenomena as a symptom dimension in psychotic disorders. Schizophr Bull 2010;36:231–8.
[13] Cohen D. Towards a valid nosography and psychopathology of catatonia in children and adolescents. Int Rev Neurobiol 2006;72:131–47.
[14] Dhossche D, Wilson C, Wachtel L. Catatonia in childhood and adolescence: implications for the DSM-5. Primary Psychiatry 2010;17:35–9.
[15] Billstedt E, Gilberg C, Gilberg C. Autism after adolescence. population-based 13- to 22-year follow-up study of 120 individuals with autism diagnosed in childhood. J Autism Dev Disord 2005;35:351–60.
[16] Cohen D, Nicolas J, Flament M, et al. Clinical relevance of chronic catatonic schizophrenia in children and adolescents: evidence from a prospective naturalistic study. Schizophr Res 2005;15:301–8.
[17] Consoli A, Boulicot V, Cornic F, Fossati P, Barbeau M, Cohen D. Moderate clinical improvement with maintenance ECT in a 17-year-old boy with intractable catatonic schizophrenia. Eur Child Adolesc Psychiatry 2009;18:250–4.
[18] Green W, Padron-Gayol M, Hardesty A, Bassiri M. Schizophrenia with childhood onset: a phenomenological study of 38 cases. J Am Acad Child Adolesc Psychiatry 1992;31:968–76.
[19] Moise FN, Petrides G. Case study: electroconvulsive therapy in adolescents. J Am Acad Child Adolesc Psychiatry 1996 Mar;35:312–8.
[20] Ohta M, Kano Y, Nagai Y. Catatonia in individuals with autism spectrum disorders in adolescence and early adulthood: a long-term prospective study. Int Rev Neurobiol 2006;72:41–54.
[21] Thakur A, Jagadheesan K, Dutta S, Sinha V. Incidence of catatonia in children and adolescents in a pediatric psychiatric clinic. Aust N Z J Psychiatry 2003;37:200–3.
[22] Wing L, Shah A. Catatonia in autistic spectrum disorders. Br J Psychiatry 2000;176:357–62.
[23] Dhossche D, Shettar S, Kumar T, Burt L. Electroconvulsive therapy for malignant catatonia in adolescence. South Med J 2009;102:1170–2.
[24] Wachtel L, Griffin M, Dhossche D, Reti I. Electroconvulsive therapy for malignant catatonia in an autistic adolescent. Autism 2010;14:349–58.
[25] Cornic F, Consoli A, Tanguy M, et al. Association of adolescent catatonia with increased mortality and morbidity: evidence from a prospective follow-up study. Schizophr Res 2009;113:233–40.
[26] Lahutte B, Cornic F, Bonnot O, et al. Multidisciplinary approach of organic catatonia in children and adolescents may improve treatment decision making. Prog Neuropsychopharmacol Biol Psychiatry 2008;32:1393–8.
[27] Marra D, Amoura Z, Soussan N, et al. Plasma exchange in patients with stuporous catatonia and systemic lupus erythematosus. Psychother Psychosom 2008;77:195–6.
[28] Perisse D, Amoura Z, Cohen D, et al. Case study: effectiveness of plasma exchange in an adolescent with systemic lupus erythematosus and catatonia. J Am Acad Child Adolesc Psychiatry 2003;42:497–9.
[29] Bailine S, Petraviciute S. Catatonia in autistic twins: role of electroconvulsive therapy. J ECT 2007;23:21–2.
[30] de Winter C, van Dijk F, Verhoeven W, Dhossche D, Stolker J. Autism and catatonia: successful treatment using lorazepam. A case study. Tijdschrift voor Psychiatrie 2007;49:257–61.
[31] Dhossche D. Catatonia in autistic disorders (brief report). J Autism Dev Disord 1998;28:329–31.
[32] Dhossche D, Reti I, Wachtel L. Catatonia and autism: a historical review, with implications for ECT. J ECT 2009;25:19–22.
[33] Dhossche D, Wing L, Ohta M, Neumarker K-J, editors. Catatonia in autism spectrum disorders. San Diego, California and London, UK: Elsevier Academic Press; 2006.
[34] Fink M, Taylor M, Ghaziuddin N. Catatonia in autistic spectrum disorders: a medical treatment algorithm. Int Rev Neurobiol 2006;72:233–44.
[35] Ghaziuddin M, Quinlan P, Ghaziuddin N. Catatonia in autism: a distinct subtype? J Intellect Disabil Res 2005;49:102–5.
[36] Ghaziuddin N, Gih D, Barbosa V, Maixner F, Ghaziuddin M. Catatonia with onset in puberty: ECT response in two autistic adolescents. J ECT, in press.
[37] Hare D, Malone C. Catatonia and autism spectrum disorders. Autism 2004;8:183–95.
[38] Kakooza-Mwesige A, Wachtel L, Dhossche D. Catatonia in autism: implications across the life span. Eur Child Adolesc Psychiatry 2008;17:327–35.
[39] Realmuto G, August G. Catatonia in autistic disorder: a sign of comorbidity or variable expression. J Autism Dev Disord 1991;21:517–28.
[40] Revuelta E, Bordet R, Piquet T, Ghawche F, Destee A, Goudemand M. Acute catatonia and neuroleptic malignant syndrome. A case of infantile psychosis. Encephale 1999;20:351–4.
[41] Wachtel L, Hermida A, Dhossche D. Maintenance electroconvulsive therapy in autistic catatonia: a case-series review. Prog Neuropsychopharmacol Biol Psychiatry 2010;34:581–7.
[42] Wachtel L, Kahng S, Dhossche D, Cascella N, Reti I. Electroconvulsive therapy for catatonia in an autistic girl. Am J Psychiatry 2008;165:329–33.
[43] Wing L, Shah A. A systematic examination of catatonic-like clinical pictures in autism spectrum disorders. Int Rev Neurobiol 2006;72:21–39.
[44] Zaw F, Bates G, Murali V, Bentham P. Catatonia, autism, and ECT. Dev Med Child Neurol 1999;41:843–5.
[45] Dhossche D, Bouman N. Catatonia in an adolescent with Prader–Willi syndrome. Ann Clin Psychiatry 1997;4:247–53.
[46] Verhoeven W, Tuinier S. Prader–Willi syndrome: atypical psychoses and motor dysfunctions. Int Rev Neurobiol 2006;72:119–30.
[47] Wachtel LE, Griffin M, Reti I. Electroconvulsive therapy in a man with autism experiencing severe depression, catatonia, and self-injury. J ECT 2010;96:70–3.
[48] Gaind G, Rosebush P, Mazurek M. Lorazepam treatment of acute and chronic catatonia in two mentally retarded brothers. J Clin Psychiatry 1994;55:20–3.
[49] Cavanna A, Robertson M, Critchley H. Catatonic signs in Gilles de la Tourette syndrome. Cogn Behav Neurol 2008;21:34–7.
[50] Dhossche D, Reti I, Shettar S, Wachtel L. Tics as signs of catatonia. J ECT, in press.
[51] Dhossche D, Bouman N. Catatonia in children and adolescents (letter). J Am Acad Child Adolesc Psychiatry 1997;36:870–1.
[52] Gjessing LR. Lithium citrate loading of a patient with periodic catatonia. Acta Psychiatr Scand 1967;43:372–5.
[53] Lee A, Glick D, Dinwiddie S. Electroconvulsive therapy in a pediatric patient with malignant catatonia and paraneoplastic limbic encephalitis. J ECT 2006;22:267–70.
[54] Slooter A, Braun K, Balk F, van Nieuwenhuizen O, van der Hoeven J. Electroconvulsive therapy of malignant catatonia in childhood. Pediatr Neurol 2005;32:190–2.
[55] Wachtel L, Baranano K, Reti I. Electroconvulsive therapy for catatonia in an adolescent with a pontine arachnoid cyst and shunted hydrocephalus. Pediatr Neurol, in press.
[56] Kinrys P, Logan K. Periodic catatonia in an adolescent (letter). J Am Acad Child Adolesc Psychiatry 2001;40:741–2.
[57] Croarkin P, Emslie G, Mayes T. Neuroleptic malignant syndrome associated with atypical antipsychotics in pediatric patients: a review of published cases. J Clin Psychiatry 2008;69:1157–65.
[58] Ghaziuddin N, Alkhouri I, Champine D, Quinlan P, Fluent T, Ghaziuddin M. ECT treatment of malignant catatonia/NMS in an adolescent: a useful lesson in delayed diagnosis and treatment. J ECT 2002;18:95–8.
[59] Neuhut R, Lindenmayer J, Silva R. Neuroleptic malignant syndrome in children and adolescents on atypical antipsychotic medication: a review. J Child Adolesc Psychopharmacol 2009;19:415–22.
[60] Steingard R, Khan A, Gonzalez A, Herzog D. Neuroleptic malignant syndrome: review of experience with children and adolescents. J Child Adolesc Psychopharmacol 1992;2:183–98.
[61] Ty E, Rothner A. Neuroleptic malignant syndrome in children and adolescents. J Child Neurol 2001;16:157–63.
[62] Rani F, Byrne P, Murray M, Carter P, Wong I. Paediatric atypical antipsychotic monitoring safety (PAMS) study: pilot study in children and adolescents in secondary- and tertiary-care settings. Drug Saf 2009;32:325–33.
[63] Tamminga C, Sirovatka P, Regier D, van Os J. Deconstructing psychosis: refining the research agenda for DSM-V. Arlington, VA: APA, Inc.; 2010.

Please cite this article in press as: Dhossche D et al. The study of pediatric catatonia supports a home of its own for catatonia in DSM-5. Med Hypotheses (2010), doi:10.1016/j.mehy.2010.07.029