

INVESTIGATING THE USE OF PACKING THERAPY IN ADOLESCENTS WITH CATATONIA:
A RETROSPECTIVE STUDY

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Abstract

Packing therapy is an adjunct symptomatic treatment used in autism and/or catatonia that has not been systematically assessed. Since 1993, we have conducted at the Pitié-Salpêtrière Hospital a prospective study on the phenomenology and psychopathology of catatonia in children and adolescents. Here, we reviewed the charts of all the catatonic patients who received packing therapy. During the study period, among the 44 patients included in the prospective series, 6 (5 males and 1 female) had packing during their stay. (i) Packing therapy appeared to be an interesting adjunct treatment in 4 patients; (ii) the main effect relied on the specific sensory/coenesthetic experience, it offered the patients who expressed and showed severe distortion in sensory and body image representation; (iii) overall tolerance and compliance were good. We conclude that packing seems to be a feasible and interesting adjunct treatment in catatonia providing that the consent could be obtained from the patient and a significant relative as a parent. Given the limitations of this study (small sample size, retrospective design, no blind assessment, confounding concurrent pharmacotherapy, no definition of responders), further clinical studies should be promoted to clarify its possible efficacy and underlying mechanism.

KeyWords: Catatonia – Psychotic Disorders – Sensory integration – Packing – Adolescent

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Introduction

Packing therapy is an adjunct treatment given by occupational therapists that is based on patient's envelopment in damp sheets for one hour sessions while he expresses coenesthetic sensations and spontaneous fantasies (Cohen et al. 1999). In a report published in the Lancet, Spinney (2007) investigated the controversy regarding the use of packing therapy in France for autism. The controversy relies on: (i) the absence of evidence-based study to support the treatment; (ii) the possible absence of free consent in individuals with poor communication skills; (iii) with respect to autism which constitutes the current main indication of packing therapy, a theoretical background that is erroneously associated to psychoanalysis. This last point being crucial as the controversies between parents' associations in the field of autism and psychodynamic

theoretical background is still vivid in France (Chamak and Cohen 2003) and abroad (Rhode 2008).

As we could not find either any evidence-based study supporting packing therapy, we would report on our own experience of packing therapy in an Inpatient Psychiatric Unit with adolescents presenting Catatonia. In our view the packing therapy is better conceived as a sensory integration approach in the sense of A.J. Ayres (2005). Sensory integration is the hierarchical organisation of the somatic sensations which serve as foundations of the individual's perceptions, behaviours and learning. The greatest potential for the development of sensory integration occurs within an adaptation response, which is a purposeful, goal-directed response to a sensory experience. Auditory, vestibular, proprioceptive, tactile and visual senses are progressively integrated as a *body percept* in which are rooted different psychosomatic functions such as the

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coordination of the two sides of the body, motor planning, activity level, attention span and emotional stability. Sensory Integration Dysfunction (SID) results in a wide variety of Learning and Developmental Disorders. Considering the poor sensory processing observed in Autism Spectrum Disorders, the SID as described by Ayres is presently viewed as a core deficit on which treatment interventions should be focused (Bauman 2005, Greenspan et al. 2008, Kloeckner et al. 2009). Its importance is also reported by individual with autism who can express themselves (Chamak et al. 2008). Apart from learning and severe developmental disorders, the SID could contribute to other clinical pictures without explaining the whole cognitive dysfunctions (Bundy 2005, Bundy et al. 2007). In that vein we propose to view Catatonia as a SID occurring in neurological diseases and severe psychiatric conditions such as Psychotic Disorders.

Although infrequent in adolescence, Catatonia is a severe condition in which several deaths have been reported (Ainsworth 1987, Dimitri 2006). This neuropsychiatric condition severely impedes the patient's functioning. Catatonic patients face huge impairments in everyday life: waking up, washing themselves, getting dressed, eating, and of course getting involved in any activity. Catatonia occurs in various psychiatric disorders, neurological diseases, intoxications and metabolic conditions (Cornie et al. 2007, Lahutte et al. 2008). Its phenomenology encompasses motor (e.g. posturing, catalepsy, waxy flexibility), behavioural (e.g. negativism, mutism, automatic compulsive movements), affective (e.g. involuntary and uncontrollable emotional reactions, affective latency, flat affect and withdrawal), and regressive symptoms (e.g. verbigeration, enuresis and encopresis, echophenomena). In adults, epidemiological studies using catatonia rating scales found that the prevalence of catatonia ranges from 7.6% to 38% among psychiatric inpatients (Taylor and Fink 2003). The syndrome is more frequent in female patients, and is usually associated with mood disorders with psychotic features (Taylor and Fink 2003). There are few studies on catatonia in the field of child and adolescent psychiatry. The prevalence of catatonia in youth psychiatric inpatients ranges from 0.6% in a prospective study in France to 17.7% in India (Cohen et al. 2005, Thakur et al. 2003). This discrepancy between these two studies suggests that cultural factors may play a role with catatonic child and adolescent inpatients. While the phenomenology and associated disorders are similar to those reported in the adult literature, findings differ with regard to the female-to-male ratio and the relative frequencies of associated disorders. Catatonia in children or adolescents is more frequent in boys (Takaoka and Takata 2003) and schizophrenia is the most frequent associated diagnosis (Cornie et al. 2007). Furthermore, a personal history of pervasive developmental disorders (PDD) may be found in young patients with catatonia (Billstedt et al. 2005, Ohta et al. 2006, Wing and Shah 2000, Kakooza-Mwesige 2008). The recommended treatments are symptomatic and include the use of sedative drugs (e.g. benzodiazepines) and electroconvulsive therapy (ECT) that offer some dramatic and rapid improvement in many cases (Taylor and Fink 2003, Caroff et al. 2004). However, treatment

of causal organic condition may be helpful on psychomotor symptoms as well. Recently, we showed that plasma exchanges (PE) could be an efficient treatment option for catatonic manifestations of systemic lupus erythematosus, avoiding the use of ECT (Marra et al. 2008). We propose in this article to consider catatonia as an acute and severe Sensory Integration Dysfunction (SID) state associated with body maps disorganization, absence of integration of proprioceptive sensations, failing in motor planning which can explain motor symptoms, whereas affective symptoms can be conceived as an ultimate product of the failure of sensory integration processes.

When catatonia does not respond to high dose of benzodiazepines, therapists facing youths with catatonia are quite armless as family are reluctant to accept ECT. Resistance to psychotropic medication (Cohen et al. 2005) or side effects (e.g. Extra-Pyramidal Syndrome and/or sedation) (Cohen et al. 1999) usually developed during the course of treatment. With respect to its theoretical groundings in sensory integration theory, packing therapy could be of interest in these clinical situations. The packing therapy is a particular treatment based on multisensory (tactile, coenesthetic and proprioceptive) stimulations. During packing therapy the patient's body is wrapped in cold wet sheets first and soon after in dry sheets. The head of the patient remains free from the wrapping, which allows for communication through visual and auditory channels. Delion hypothesized that the packing intervention saturates tactile feelings and facilitates contact through visual and auditory senses (Delion 2007). Following A.J. Ayres who defines sensory integration therapy as a 'therapy which involves the whole body, all the senses, and the entire brain', we would assume that packing therapy rely on a sensory integration approach.

In the present study, we retrospectively reviewed therapeutic data on all patients who received packing sessions in the context of an ongoing prospective study on Catatonia in adolescents (Cohen et al. 1999, 2005). We focused first on the therapeutic content and clinical variables at admission and discharge from hospital, second on both the clinician's and the patient's view regarding packing interventions. This study aims at illustrating the feasibility of packing therapies in Catatonia, providing clinical data regarding the impact of packing therapy on the course of the disorder and narratives from the clinician and the patient undergoing the therapy.

Method

Participants

The sample includes six patients who received packing treatment as part of a naturalistic prospective study on Catatonia conducted since 1993, in the Department of Child and Adolescent Psychiatry of a University Teaching Hospital (GH Pitié-Salpêtrière, Paris). Every admission is screened for catatonic symptoms. When the presence of Catatonia is supported by the psychiatric evaluation and the benefit from psychotropic medication insufficient, packing therapy is proposed as an adjunct/alternative treatment for those

who are not eligible or refuse ECT. The present sample (**table 1**) regroups five boys and one girl (mean age: 15.16 years) presenting either a Psychotic Disorder (n=5) or a Major Depressive Disorder with psychotic features (n=1). Two had a psychiatric history positive for Pervasive Developmental Disorder (PDD).

Intervention: packing therapy

The therapy is aiming at restoring sensory integration and building proper body representations. The overall treatment encompasses a series of twice-a-week sessions over a minimum of one-month period. Usually lasting one hour each session can be expanded until two hours depending on the patient's response. Durations in the present sample are displayed in **table 1**. Sessions are conducted under the supervision of a *psychomotricien*¹ and gather at least two members of the patient's care team. At the beginning of the session the patient is first wrapped up in cold damp sheets (cold phase) and covered up with a rescue and a dry blanket. Afterward the body spontaneously warms up (warm phase). The patient is then invited to freely express his feelings, coenesthetic sensations and somatic fantasies. Cardiac and Respiratory Frequencies as well as Blood Pressure are monitored during the session to detect cardio-vascular adverse effects and/or adverse autonomous reactions. At the end of the session, a brief moment of drawing or modeling with clay is proposed in order to provide non-verbal avenues to express feelings and explore body representations. Throughout the session, the patient's comments and relevant observations from the clinicians (e.g. clinical signs, body image, and coenesthetic sensations) are carefully recorded by one of the participants. The patient's verbalizations and projective material are later discussed at the meeting of the clinical team including a psychologist, the occupational therapist and the treating psychiatrist.

Psychiatric and somatic evaluation

Systematic assessment within the first week of admission included (i) socio-demographic data (age, sex, origin and socio-economic status of the family); (ii) personal and family history of psychiatric/somatic disorders acquired through a semi-structured interview (detailed in Taieb et al. 2002); (iii) clinical assessment jointly conducted by a child psychiatrist and a pediatrician. The clinical diagnosis of Catatonia requires the presence of at least two catatonic motor signs (catalepsy, stupor, posturing, waxy flexibility, staring, negativism, stereotypies, psychomotor excitement, automatic compulsive movements, grimacing, echopraxia, muscular rigidity), or one catatonic motor sign combined with a non-motor catatonic symptom

(social withdrawal, mutism, mannerism, echolalia, incontinence, verbigeration (i.e., meaningless and stereotyped repetition of words), schizophasia (scrambled speech), acrocyanosis (cyanosis of the extremities), and refusal to eat) indicative of severe impairment in behavioral and emotional functioning (Cohen et al. 1999, 2005); (iv) type of onset of catatonia (< 10 days = acute; >10 days = insidious) and duration of the episode; (v) evaluation of the presence and severity of catatonic symptoms as well as the overall functioning with clinician-rated scales. (vi) At entry and discharge from the hospital, patient was scored on the Clinical Global Impression-Severity (CGI-S) (Guy 1976) and the Global Assessment of Functioning (GAF) scales. For description of the phenomenology of catatonic signs, we used a modified version of the 14 items Bush-Francis Catatonia Rating Scale (Bush et al. 1996), to which were added the following six symptoms taken from Ey's earlier description (Ey 1950): catalepsy, refusal to eat, incontinence, acrocyanosis, schizophasia and automatic compulsive movements (range scores: 0-60) (Cohen et al. 2005). As the study is retrospective, there was no a priori definition of responders.

Other investigations include repeated somatic and psychiatric evaluations. Besides Catatonia, psychiatric diagnoses were carried out through the team-consensus best estimate procedure (Klein et al. 1994) with respect to the DSM-IV criteria (APA 2003) the team-consensus included the child psychiatrist in charge of the patient and two other psychiatrists experienced into child and adolescent psychiatry (DC, DP, AC). Given the frequency of negativism, mutism or stupor in instances of such severe clinical presentations, semi-structured diagnostic interviews (such as Kiddie-SADS) were not helpful. Additional investigations always included routine hematological tests, electroencephalography, and neuroimaging. Depending on both psychiatric and medical examinations, or in cases of resistance to pharmacotherapy, cerebrospinal fluid exploration and appropriate screening tests for the known metabolic causes associated with the syndrome were performed.

Procedure

Data were prospectively collected on consecutive admissions for Catatonia (n=44). Within that sample, participants who received packing therapy were identified and their chart retrospectively reviewed on data pertaining to the packing therapy sessions and additional data (e.g. duration of hospitalization). Selection criteria were as follows: 1) Patient had to present with Catatonia as per diagnostic criteria described above; 2) Patient had to be resistant to or not eligible for other medication or therapeutics, and consented to receive packing therapy. Psychiatric assessments as well as the clinician-rated scales were administered by one of the three psychiatrists experienced in adolescence and Catatonia (DC, DP, AC). Medical conditions were ruled out. As the medical recommendation to use packing therapy was presented to the family, the process of the therapy as well as the benefits and possible adverse effects were explained to the adolescent and his/her parent(s). According to the Hospital Ethics regulation, the patient's parent consent

¹ a *psychomotricien* is a therapist holding a French diploma in *psychomotricité* which is a specialized training in psychomotor disturbances within the Occupational Therapy course.

was obtained in all cases. In all cases but one who had no access to verbal language (case 1), we obtained the patient's consent to the therapy. In one occurrence (case 1) only the assent of the patient's parents was obtained. Of note, case 2 (and his parents) refused ECT whereas a change in antipsychotic medication combined with packing therapy was accepted.

Results

All participants presented with a severe psychiatric condition evidenced by the severity scores (GAF: global assessment functioning; CGI: clinical global impression; CRS: Bush-Francis Catatonia Rating Scale), duration of hospitalisation (range: 3 to 12 months), and number of previous medication trials without reaching sufficient clinical improvement (**table 1**). Following packing therapy, no side effects were reported. As indicated in **table 1**, all 6 patients improved their clinical status severity during hospitalization whatever the treatment that included 3 to 18 sessions of packing.

According to the clinician's view, packing together with current psychotropic drugs appeared to be helpful in 4 patients including the 2 patients with a history of PDD. For case 6, the response was uncertain as he only received 3 sessions and had a moderate overall improvement. For case 3, there was no response to any treatment except ECT that allowed the patient to be discharged from the hospital. Clinicians also reported interesting improvements regarding body image (N=2), sensory integration such as increase in cold sensitivity (N=5) and tonic motor status during sessions (N=4).

In most cases, patients experienced a parallel to that of clinicians (narratives are reported in **table 1**). During the first sessions, a participant (case 2) reported his body to be "a piece of metal" and was sleeping during the "cold time" of the session with no feeling of the cold sensation. Clinical improvement paralleled improvement of cold sensitivity with relaxation/sleeping during the warm time of the session. Finally, when he reached substantial improvement, the patient asked to "stop this unpleasant treatment" as he was feeling again the cold sensation at the beginning of the session. Case 5 showed a similar course and even found "sessions pleasant and too short". Case 4 stated that packing helped her "to feel more relaxed from the neck to the feet", and case 6 reported that "packing was a moment of resting, and relaxation". Finally, only case 3 expressed unpleasant experience during packing. This patient was the one that did not show improvement with packing and/or psychotropic medication. He only improved after a series of 15 ECT.

Discussion

Before discussing the results, one should keep in mind the limitations of the study: (1) despite a prospective recruitment, the data regarding packing therapy were retrospectively collected; (2) the sample size was small; (3) the evaluation of the severity was not conducted blindly to the diagnosis; (4) no comparison were made to a control series; (5)

confounding concurrent pharmacotherapy were used in all patients (see **table 1**); (6) given the retrospective design, no *a priori* definition of responders was used. Despite these limitations, our data which are the first case series published on packing therapy to our best knowledge, tend to support the feasibility of this therapy in adolescents presenting with a severe psychotic condition associated with poor communication skills. In addition, without directly addressing Spinney's concerns dealing with autism (Spinney 2007), our series indicate that the packing therapy may be an interesting adjunct treatment in youths presenting Catatonia. In keeping with the clinicians' observations and the patients' narratives, a combined treatment with packing therapy and psychotropic medications, including antipsychotics, clearly appeared to provide symptomatic and subjective relief to the patient. With respect to Spinney's concern, stating that packing may be harmful in patients with autism who can not always express themselves, we were able to carefully collect the patient as well as the parents' consent and document the patient's subjective experience. The treatment was rather well accepted in adolescents with Catatonia. All of them accepted the treatment and could express themselves on the treatment (except the patient without language). In 5 out 6 patients, treatment was positively viewed and a decrease in anxiety was experienced.

Considering the phenomenology of the syndrome described in the introduction, it is therefore extremely difficult to identify the subjective feelings experienced by a catatonic patient when negativism is high. For those who have an acute form of Catatonia (e.g. some form of psychotic depression) (Cohen et al. 1997, Northoff et al. 1999), it is possible to retrospectively ask patients about such subjective experience. For those who have a chronic form of catatonia (e.g. some forms of catatonic schizophrenia) (Cohen et al. 1999), documenting the subjective experience requires that the patient do not show extreme and enduring mutism or negativism, and is able to later engage in a confident relationship with the treating team. A few studies (Rosebus and Mazurek 1999, Cohen 2006, Northoff et al. 1998) shed light on the subjective experience of catatonic patients. First, akinetic patients with catatonia appear unable to experience pain or fatigue despite prolonged posturing. This statement is supported by the occurrence of skin injury lesions as a consequence of Catatonia, even in young patients (Cohen et al. 1999). Second, they appear unaware of the objective position of their body, or of the consequences of their movements. Third, most of them report intense and uncontrollable emotions, including one patient who had a blockade of his will with contradictory and ambivalent thoughts. Fourth, all patients of Northoff et al. series (Northoff et al. 1998) 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 Rosebus and Mazurek (1999). We do share the same experience with young patients except when a history of autism with no language does not permit retrospective psychological investigation (Cohen 2006). Similarly, except when catatonia is associated with a neurological disorder, catatonic patients do not have abnormal neurological examination (Cohen et al. 2005, Northoff et al. 1999,

Table 1. Clinical characteristics, treatment, and clinician's and patient's view of 6 consecutive adolescents who had packing as adjunct therapy in catatonia

CLINICAL CHARACTERISTICS												
	Case 1		Case 2		Case 3		Case 4		Case 5		Case 6	
Age	17		13		14		14		17		16	
Gender	M		M		M		F		M		M	
History	Autistic Dis.		Asperger Dis.		No		No		No		No	
DSM-IV diagnosis	Psycho-NOS		SCZ		SCZ		MDE		SCZ		Psycho-NOS	
Catatonia duration*	> 12		> 12		> 12		12		12		6	
Inpatient duration*	3		5.5		12		7		5		4	
Clinical variables	ADM	DIS	ADM	DIS	ADM	DIS	ADM	DIS	ADM	DIS	ADM	DIS
CGI-S	7	6	7	6	7	5	7	4	7	4	6	6
GAF	10	40	15	40	10	40	11	60	23	55	25	40
CRS	30	15	29	9	20	8	25	0	17	4	16	9
TREATMENT REGIMEN												
Inefficient treatment	fluoxetine; sertraline; risperidone; amisulpride; haloperidol		clomipramine; cyamemazine; chlorpromazine; thioridazine; clorazepate; lorazepam		paroxetine; sertraline; amisulpride; thioridazine; haloperidol; olanzapine; flupentixol; lorazepam		clomipramine; clorazepate; prazepam		fluoxetine; lorazepam		Risperidone	
Efficient treatment	clomipramine; olanzapine; lorazepam; packing		amisulpride; packing		ECT (N=15); olanzapine		fluoxetine; amisulpride; lithium; packing		risperidone; packing		fluoxetine; lorazepam; packing?	
PACKING												
Number of sessions	18		18		16**		13		15		3	
Duration*	2.5		3.5		3		3		3		0.75	
Overall judgment efficacy	Yes		Yes		No		Yes		Yes		No	
Clinician's view	Improvement of cold sensitivity		Improvement of cold sensitivity Temporary improvement of catatonia just after sessions Discordant body/sensory representation		Did not like treatment No effect on catatonia and other symptoms		Improvement of cold sensitivity More and more relaxed through sessions		Improvement of cold sensitivity Improvement of body image in drawings More and more relaxed through sessions		Slight improvement of cold sensitivity. Very relaxed during first session.	
Patient's view	Severe mutism prevent the patient from expressing his feelings First word during packing		Gave consent to packing Slept during session Clinical improvement paralleled improvement of cold sensitivity and request to stop		Gave consent to packing Refused several times to be wrapped in the sheets claiming it was too freezing Finally asked to stop this treatment		Gave consent to packing Asked for more sessions Felt more relaxed and said to feel better some parts of her body		Gave consent to packing Felt his muscles relaxed Found the sessions pleasant and too short		Gave consent to packing Found sessions very pleasant, painless and relaxing	
*(months); **only 10 sessions were completed; ADM: admission; DIS: discharge; M: male; F: female; MDE: major depressive episode with psychotic and catatonic features; SCZ: early-onset schizophrenia; Psycho-NOS: psychosis not otherwise specified; GAF: global assessment functioning; CGI: clinical global impression; CRS: Bush-Francis Catatonia Rating Scale; ECT: electro-convulsive therapy.												

Rosebush and Mazurek 1999). Neurological functioning is usually preserved in catatonic patients. Catatonic symptoms should be regarded as functional and understood at the level of the subjective experience resulting in catatonic motor dysfunction.

The patients' narratives reported during packing sessions support the idea that catatonic experiences imply severe distortion in sensory/coenesthetic inputs and body image representations (see **table 1**). Classical/normal coenesthetic sensations during packing sessions is to feel cold at the beginning of the session because of the cold wet sheets and then experience a progressive warming up of the body leading to a relaxation effect

and sometimes sleep. Body representation is reinforced by the fact that the whole body is wrapped up and stimulated by the wet sheet. We can hypothesize that packing provides to the patient a new coenesthetic experience together with a holding effect that helped, as Delion stated (2007), to "combine the body and the image of the body" and "to reinforce children's consciousness of their body limits". Helping patients mastering their sensory processing issues packing therapy leads catatonic patients to recover their motivation to move ahead and participate in daily activities. The observed increase in GAF scores might reflect this subsequent improvement. In the same way,

physiological experiments have shown that vestibular irrigation of the ear with cold water may reverse anosognosia, somatoparaphrenia and neglect in neurological patients with right parietal lobe lesions (Cappa et al. 1987).

We conclude that, in the case of Catatonia, a syndrome which associates motor and psychic symptoms, packing therapy operating as a sensori-integration approach, seems to be a feasible and interesting adjunct treatment. Consent has to be collected from the patient and from the parent(s). Although speculative, patients' experience and clinicians' view support the idea that packing may improve body image and integration through a unique sensory/motor experience, that we can figure as a sensory 're-integration'. However, further clinical studies should be promoted in order to clarify its possible efficacy and underlying mechanism.

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