Lorazepam, fluoxetine and packing therapy in an adolescent with pervasive developmental disorder and catatonia

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Abstract

Packing therapy is an adjunct symptomatic treatment used for autism and/or catatonia. Here, we report the case of a 15-year-old boy with pervasive developmental disorder who developed catatonia. At admission, catatonic symptoms were severe and the patient required a feeding tube. Lorazepam up to 15 mg/day moderately improved the catatonic symptoms. On day 36 we added fluoxetine and on day 62 we added packing therapy (twice per week, 10 sessions). After three packing sessions, the patient showed a significant clinical improvement (P < 0.001). At discharge (day 96), he was able to return to his special education program. Although we do not consider packing as a psychodynamic treatment, this case challenges the concept of embodied self that has opened new perspectives on a dialogue between psychoanalysis and neuroscience. Indeed, better body representation following packing sessions, as shown in the patient’s drawing, paralleled clinical improvement, and supports the concept of embodied self. This concept may serve as a link between psychoanalysis and attachment theory, developmental psychology with the early description of “sense of self”, and cognitive neurosciences that more and more support the concept of embodied cognition. Further clinical studies are necessary to clarify the efficacy and underlying mechanism of packing treatment and to understand how patient’s experience may illustrate the concept of embodied self.

1. Introduction

The concept of embodied self has open new perspectives on a dialogue between psychoanalysis and neurosciences (Fonagy and Target, 2007; Gallese, 2007). In this report, we aimed to discuss how packing therapy that was given to an adolescent with autism and catatonia, and the clinical response over time, illustrate the concept of embodied self. Before summarizing and discussing the case report, a brief overview on packing therapy, on sensory integration and catatonia is warranted.

1.1. Packing therapy

Packing therapy is based on multisensory (tactile, c esthanesic and proprioceptive) stimulations. Ross et al. (1988) conducted a national survey which demonstrated that it was rarely used in modern American psychiatry. By reviewing its use in 46 hospitalized psychiatric patients, they concluded that the treatment was safe and had interesting and useful effects that go beyond the concept of simple restraint.

The overall treatment encompasses a series of two sessions per week over a minimum one-month period. Usually, each session lasts one hour; however, the session time can be expanded to two hours depending on the patient’s response. Sessions are conducted under the supervision of a psychomotricien1 and at least two members of the patient’s care team are present (Cohen et al., 2009; Delion, 2007). First, the patient is wrapped in damp sheets (cold phase). Then, the patient is covered up with a rescue cover and a dry blanket, and the body spontaneously warms up (warm phase). The head of the patient remains free from the wrapping, which allows for communication through visual and auditory channels. Cardiac and respiratory frequencies and blood pressure are monitored before and after the session to detect adverse cardio-vas-

1 A psychomotricien is a therapist holding a French diploma in psychomotricité which is specialized training in psychomotor disturbances within the Occupational Therapy course.
cular effects and/or adverse autonomous reactions. At the end of the session, the patient is asked to draw or model with clay in order to provide non-verbal avenues to express feelings and explore body representations. Throughout the session, the patient's comments and clinicians' relevant observations (e.g. clinical signs, body image, and cenesthesis sensations) are carefully recorded by one of the participants (Cohen et al., 2009). Packing is used as an adjunct treatment in two main indications: catatonia (Chamak and Cohen, 2009) and behavioral disturbances occurring in autism or mental retardation (Loby et al., submitted for publication; Goeb et al., 2009). Recent controversies have emerged based on: (i) the absence of an evidence-based study to support the treatment; (ii) the possible absence of free consent in individuals with poor communication skills; and (iii) erroneous association of the theoretical background of packing with psychoanalysis, despite the fact patient's experience of packing may contribute to the psychodynamic metapsychology of the self (Delion, 2007). This final point is crucial, as there is strong disagreement between parents associations and psychodynamic theory in the field of autism in France (Chamak and Cohen, 2003) and abroad (Rhode, 2008).

1.2. Is packing a sensory-integration approach?

In our view packing therapy is better understood as a sensory-integration approach as described by Ayres (2005) or Bullinger (Kloekner et al., 2009). Sensory integration is the hierarchical organization of the somatic sensations that serve as foundations for the individual's perceptions, behaviors 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, and are rooted in different psychosomatic functions such as the 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 developmental disorders (Bundy, 2005; Bundy et al., 2007). Considering the poor sensory processing observed in Autism Spectrum Disorders, SID is viewed as a core deficit on which treatment interventions should be focused (Bauman, 2005; Greespan et al., 2008; Kloekner et al., 2009). Individuals with autism who can express themselves have also reported the importance of sensory processing (Chamak et al., 2008). In addition to learning disorders and severe developmental disorders, SID may contribute to other clinical symptoms such as catatonia without entirely explaining the cognitive dysfunctions (Cohen et al., 2009).

1.3. Catatonic syndrome in adolescents

Although infrequent in adolescence, catatonia is a severe condition; several deaths have been reported (Ainsworth, 1987; Dimitri et al., 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 attempting any activity (Cormic et al., in press). Catatonia occurs in various psychiatric disorders, neurological diseases, intoxications and metabolic conditions (Takaoka and Takata, 2003; Cormic et al., 2007; Lahutte et al., 2008). Its phenomenology encompasses motor (e.g., posturing, catalepsy, waxy flexibility), behavioral (e.g., negativism, mutism, automatic compulsive movements), affective (e.g., involuntary and uncontrollable emotional reactions, affective latency, flat affect, withdrawal), and regressive symptoms (e.g., verbigeration, enuresis and encopresis, echopraxia) (Northoff et al., 1999). Catatonia can occur in young people with history of pervasive developmental disorders (PDD). In these cases, morbidity is often extremely severe and treatment is difficult (Billstedt et al., 2005; Ohta et al., 2006; Wing and Shah, 2000; Kakooza-Mwesige et al., 2008). The recommended treatments are symptomatic and include the use of sedative drugs (e.g., high doses of benzodiazepines) and electroconvulsive therapy (ECT); these treatments offer dramatic and rapid improvement in many cases (Taylor and Fink, 2003; Caroff et al., 2004; Wachtel et al., 2008). But cases with poor improvement have been reported as well (Consoli et al., 2009). When available, treatment of any associated medical condition is required as well (Cormic et al., 2007; Marra et al., 2008).

Catatonia can be viewed as an acute and severe sensory integration dysfunction (SID) state associated with body map disorganization, absence of integration of proprioceptive sensations and failure in motor planning that can explain motor symptoms; in addition, affective symptoms can be viewed as the ultimate product of a failure of sensory integration processes (Cohen et al., 2009). Considering this, we developed a packing therapy for treatment of catatonia that may be used when patients do not respond to high doses of benzodiazepines or when family members are reluctant to accept ECT. In a prospective study on youth catatonia from 1993 to 2007 (Cohen et al., 2005; Cormic et al., 2009), among the 44 patients recruited in a university setting, six adolescents (five males and one female) experienced packing during their stay, including two patients with PDD. Packing therapy appeared to be an effective adjunct treatment in four patients who also received psychotropic medications; overall tolerance and compliance were good (Cohen et al., 2009). However, we failed to find similar case descriptions in the literature.

Here, we report the case of a 15-year-old adolescent (named John) with PDD who developed catatonia by early adolescence and showed a dramatic improvement after adjunction of fluoxetine, packing and lorazepam. This case is noteworthy given (i) the severity of the patient's symptoms at admission, such as the patient need for tube feeding, (ii) the careful monitoring of catatonic symptoms, (iii) the parents consent to record packing sessions on video before and after improvement (available on website); and (iv) the dramatic improvement of John's body representation – as evidenced in drawings – that paralleled clinical improvement.

2. Case report

2.1. Family history

There is a family history of bipolar disorder in John's maternal grandfather, who received ECT and mood stabilizers and recovered. John's paternal grandmother showed symptoms of psychiatric disorders, but diagnosis was not available.

2.2. Early developmental and clinical history

John was the second child of unrelated parents. At birth, John weighed 3.47 kg and was 51 cm long with a 36 cm head circumference. His early development was remarkable. He walked at 16 months of age, and spoke his first words at 15 months and his first sentences between 18 and 24 months of age.

His social abilities were normal. At 30 months of age he had a dramatic regression of language simultaneously with the appearance of social withdrawal and stereotypes. At four years of age, investigations confirmed the diagnosis of disintegrative developmental disorder. Landau–Kleffner syndrome or other epileptic
encephalopathy was ruled out. The following exams yielded normal results: (i) genetic tests such as high resolution loading karyotype, search of the fragile X mutation by DNA assay and search of a 22q13.3 deletion by Fluorescence In Situ Hybridization (FISH); (ii) metabolic tests for organic acid and amino acid chromatography, lactate, uric acid, pyruvic acid in blood, mucopolysaccharides, glycosaminoglycan, oligosaccharides, guanidinoacetic acid and Bratton–Marshall test in urine; and (iii) neuro-imaging exams. Table 1 summarizes John’s ADI-R scores and cognitive abilities. John received outpatient special education, speech therapy and occupational therapy and attended a specific classroom for PDD individuals until adolescence. He never developed expressive language, but was able to communicate using pictograms and sign language. He was able to participate in leisure activities.

2.3. Catatonia onset

By the age of twelve, John started to show catatonic syndrome. His parents described a progressive onset and a moderate improvement of catatonic symptoms during summertime during the first year of symptoms. However, John’s condition worsened and, after a loss of 8 kg in two months during the summer of 2008, he was referred to a pediatric unit where a feeding tube was ordered. John’s psychiatrist prescribed sertraline (25 mg/day) for two months, and John showed some improvements. However, the treatment was stopped after a fainting fit. John’s mother had to stop working to take care of her child, who stayed at home with enteral nutrition for the next six months. At admission, John’s catatonic syndrome was severe, and he had malnutrition and skin injury lesions. John weighed only 39 kg despite tube feeding. Table 1 summarizes John’s clinical characteristics at admission and discharge, and lists etiological investigations conducted during his stay. Of note, all these investigations were unremarkable except decreased thyroid hormone and vitamin D levels at admission. These values entered normal ranges after administration of nutrition.

2.4. Intervention: lorazepam, fluoxetine and packing therapy

Daily treatment is summarized in Fig. 1. The modified-Bush-Francis Catatonia rating scale (CRS) was used to monitor symptoms (Bush et al., 1996; Cohen et al., 1999). As shown in Fig. 1, lorazepam up to 15 mg/day only improved symptoms moderately (12.5% decrease of CRS scores from day 1 to day 36). We decided to introduce fluoxetine (20 mg/day) by day 36, given (i) the absence of organic diagnosis despite in depth search; (ii) John’s parents feeling that their son showed depressed mood by early adolescence; and (iii) the brief improvement under sertraline. Improvement was still moderate (14.3% decrease of CRS scores from day 36 to 62) but sufficient enough to stop tube feeding by day 46. John’s parents refused ECT as a secondary treatment option, most likely due to irrational fears involving the treatment. They easily accepted packing when it was presented as a treatment option, despite the absence of evidence-based data. Packing started at day 62. After the third session of packing, John’s symptoms improved substantially and he started to manifest his joy with the

Table 1
Clinical characteristics and etiological assessment during inpatient stay.

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<tbody>
<tr>
<td>CGI-Severity/improvement</td>
<td>7/Not adapted</td>
<td>6/2</td>
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<tr>
<td>GAF</td>
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Fig. 1. Course of catatonia rating scale score according to time and treatment given of note, olanzapine and risperidone were only given once due to adverse reactions (sedation). Catatonia scores significantly decreased more quickly after day 74 (A slopes = −0.26; t = −7.091, P < 0.001). No further improvement in significance was obtained by changing the a priori hypothesis to day 73 (t = −6.99, P < 0.001) or day 75 (t = −7.051, P < 0.001).

Fig. 1

2.5. Follow-up

John continued to have packing during the following 4 weeks, as an outpatient, once a week. Then, he exhibited progressive relapse of catatonic symptoms, despite he was maintained under the same psychotropic regimen. At 6-month follow-up, he had CRS scores (29–32) back to the acute phase, despite a new increase of lorazepam. Parents accept ECT, and John was hospitalized. He had 9 sessions of bilateral ECT (3 sessions then 2 sessions per week because of prolonged seizure). He improved substantially with CRS scores at 15 and was discharged with a monthly maintenance ECT protocol.

3. Discussion

3.1. Therapeutic approach

Therapeutic approaches for catatonia are mainly symptomatic. It is recommended to use high dosage of benzodiazepines, and to perform electroconvulsive therapy (ECT) in cases of resistance or life-threatening condition (Taylor and Fink, 2003; Wachtel et al., 2008). In the case of patient John, benzodiazepines were only moderately efficient despite high doses (lorazepam up to 15 mg/day in a 40 kg-subject), and ECT was not considered because the parents refused this option after two months of hospitalization. The timing of catatonia improvement (see Fig. 1) is compatible with the effect of fluoxetine on mood disorders, as the change in CRS score slope appeared around day 75 of treatment, which was 40 days after fluoxetine was given. The relapse after stopping packing, while John had still fluoxetine, and the timing also support a possible adjunct effect of packing therapy that started on day 62.

Although generalization is not possible with a single case and no blind evaluation, this case supports the feasibility of packing therapy in adolescents presenting severe catatonic condition associated with poor communication skills. Two other cases of PDD and catatonia were published in a brief retrospective study (Cohen et al., 2009), in which the two patients also improved with an association of psychotropic medication and packing therapy (18 sessions in total for each patient). In keeping with the clinicians observations and the patients narratives, the combined treatment clearly appeared to provide symptomatic and subjective relief to the patients (Cohen et al., 2009).

With respect to ethical concerns (Spinney, 2007; Delion, 2010) that packing may be harmful in patients with autism who cannot always express themselves, Cohen et al. (2009) were able to collect the patients as well as the parents consent and document the patients subjective experience. In this study (Cohen et al., 2009), five out six patients, treatment was positively viewed and a decrease in anxiety was experienced. Here, John accepted packing easily and was even able to express some joy after a few sessions. This was also the case for all patients in the previous study. Of note, despite the staff experience with ECT (Cohen et al., 2000; Consoli et al., 2009) John's parents favoured the use of packing in addition to antidepressant treatment. They also accepted the use of John's video for educational and research purposes. At relapse only, parents accepted ECT.

3.2. Does the current case support the sensory integration hypothesis and the concept of embodied self?

Considering the phenomenology of the syndrome, it is extremely difficult to identify the subjective feelings experienced by a catatonic patient when negativism is high. A few studies (Cohen, 2006; Northoff et al., 1998; Rosebush and Mazurek, 1999) documented the subjective experience of catatonic patients who had no previous history of PDD. First, akinetic patients with catatonia appear unable to experience pain or fatigue despite prolonged posturing (of note John had skin lesion secondary to immobility). Second, akinetic patients appear unaware of the objective position...
of their bodies or of the consequences of their movements. Third, most patients report intense and uncontrollable emotions, including patients who had a blockade of their will with contradictory and ambivalent thoughts. Fourth, patients usually remember very well the caregivers who treated them at admission, which confirms that catatonic patients have no major deficit in memory and/or general awareness (Cohen et al., 1997; Northoff et al., 1998; Rosebush and Mazurek, 1999). We observed similar experiences in young patients, except when a history of autism with no language prohibited retrospective psychological investigation. Similarly, except when catatonia is associated with a neurological disorder, catatonic patients exhibit normal neurological function (Cornic et al., 2007; Northoff et al., 1999; Rosebush and Mazurek, 1999). Catatonic symptoms should be regarded as functional and understood at the level of the subjective experience resulting in catatonic motor dysfunction.

In our previous study, the patients narratives reported during packing sessions supported the hypothesis that catatonic experiences induce severe distortion in sensory/cenesthesic inputs and body image representations (Cohen et al., 2009). Classical/normal cenesthesic sensations during packing sessions are to feel cold at the beginning of the session and then to 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 and the therapist’s massage. Instead, most catatonic patients did not report cold sensitivity at the beginning of the sessions, and showed discordant body/sensory representations. This sensory functioning was restored with improvement of catatonia observed simultaneously during the packing sessions (Cohen et al., 2009). This was also the case for John who was able to be quiet, even sleeping, showing no cold sensitivity at first sessions of packing (see video of session 2 on the website).

In terms of therapeutic effect, we first hypothesize that packing provides the patient a new cenesthesic experience, a holding effect as well as a sensory-integrative effect that helped, as Delion (2007) stated, to “combine the body and the image of the body” and “to reinforce children’s consciousness of their body limits”. Fig. 2 shows John’s drawing after packing. Interestingly, body representations only appeared at session 12 with the representation of hand, and an attempt to draw a man at session 16. This might reflect better body representation. 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). Second, packing also has a powerful relaxing effect. Typically developing individuals usually sleep during the warm time of the session. This relaxing effect comes from the warming up of the body but also from the body pressure due to wrapping. The relaxing effect of body pressure has been explained by Temple Grandin who still uses a self-made machine to produce this effect on her own body (Grandin, 1986). In sum, helping patients master their sensory processing issues through packing therapy allows catatonic patients to recover their motivation to progress and participate in daily activities. The observed decrease in John’s CRS scores might reflect this subsequent improvement.

Although we do not consider packing as a psychodynamic treatment, the relaxing effect of body pressure, together with the better body representation that paralleled clinical improvement and packing sessions, may illustrate the concept of embodied self. Despite some controversies, this concept may serve as a link between psychoanalysis and (i) attachment theory through speculations about the nature of language (Fonagy and Target, 2007); (ii) developmental psychology with the early description of “sense of self” (Stern, 1998); (iii) cognitive neurosciences that more and more support the concept of embodied cognition (e.g., Gallese, 2007).

4. Conclusion

We conclude that, in the case of PDD and catatonia, the sensory-integration approach of packing therapy is a possible and effective adjunct treatment in cases of resistance to high dosage benzodiazepine and as an alternative to ECT. Consent for packing therapy must be collected from patients and parents. Further clinical studies are necessary to clarify the efficacy and underlying mechanism of packing treatment and to understand how patient’s experience may illustrate the concept of embodied self.
Acknowledgements

The authors thank John’s parents for their implication in the treatment, for trusting us during the prolonged inpatient treatment, and for allowing the publication of John’s case with videos. The authors also thank Bruno Falissard, MD, PhD for his advice regarding statistics.

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