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 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.

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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, 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, cutaneous 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, 2006). 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

1 A psychomotricien is a therapist holding a French diploma in psychomotricité which is specialized training in psychomotor disturbances within the Occupational Therapy course.
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; Cornic 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 website2); 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 unremarkable. He walked at 10 months of age, but diagnosis was not available.

His early development was unremarkable. He walked at 14 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 4 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

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2 The current case is available on the University Pierre et Marie Curie website: http://www.chups.jussieu.fr/en-ligne/index.html press psych under the pdf presentation entitled “Catatonia-Packing”. ID: PACKING and PASS WORD: PHYSIOLOGY are required to watch the videos.
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.

Table 1

<table>
<thead>
<tr>
<th>Clinical characteristics</th>
<th>Admission</th>
<th>Discharge</th>
</tr>
</thead>
<tbody>
<tr>
<td>CGI-Severity/improvement</td>
<td>7/Not adapted</td>
<td>6/2</td>
</tr>
<tr>
<td>GAF</td>
<td>30</td>
<td>36.5</td>
</tr>
<tr>
<td>CARS</td>
<td>56</td>
<td>32</td>
</tr>
<tr>
<td>CRS</td>
<td>32</td>
<td>11</td>
</tr>
<tr>
<td>Catatonic symptoms</td>
<td>Catalepsy, stupor, posturing, waxy flexibility, staring, negativism, rigidity, withdrawal, mutism, echopraxy, incontinence, acrocyanosis, refusal to eat</td>
<td>Catalepsy, stupor, posturing, waxy flexibility, negativism, withdrawal, mutism, echopraxy, refusal to eat</td>
</tr>
</tbody>
</table>

Etiological assessment

<table>
<thead>
<tr>
<th>Blood tests</th>
<th>Hemoglobin, blood cell count, blood chemistry (electrolytes, glucose, creatinine, blood urea, calcium, phosphate, magnesium level, liver function tests), erythrocyte sedimentation rate, ammonia, homocysteinaemia, copper, ceruleoplasmin, organic acid chromatography, amino acid chromatography</th>
</tr>
</thead>
<tbody>
<tr>
<td>Cerebral spinal fluid</td>
<td>Protein, glucose, cells, cultures, PCR test for Whippel's disease, serotonin and metabolites, folate</td>
</tr>
<tr>
<td>Urine</td>
<td>Copper, glycosaminoglycans</td>
</tr>
<tr>
<td>Immunological investigations</td>
<td>Antinuclear antibodies, complement fraction C3</td>
</tr>
<tr>
<td>Neurophysiological testing</td>
<td>Sleeping electroencephalography, electro-auditive potentials</td>
</tr>
<tr>
<td>Imaging</td>
<td>Brain MRI, brain spectroMRI in search of creatine transporter deficiency, abdominal ultrasonography</td>
</tr>
<tr>
<td>Genetic testing</td>
<td>Karyotype using Genome Wide Scan microarrays, search of mutations in ARX, MECP2, PABT1 genes</td>
</tr>
</tbody>
</table>

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 treatment. Lorazepam was tapered progressively to 7.5 mg/day. At discharge on day 96, John's parents expressed that they felt their son was much better.
child was in the same condition as he was before catatonia, and they wanted John to continue outpatient treatment. John still had catatonic symptoms (Table 1), but they were of less severity. He was able to go back to his special education program for the first time in two years. Overall, at discharge, the decrease of CRS scores reached 65%. For descriptive purposes, videos of packing session 2 and 13 are available on the UPMC website.

2.5. Follow-up

John continued to have packing during the following 4 weeks, as an outpatient, once a week. 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 discharge 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 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., 2007 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 posture (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 (Cormic 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/cenesthetic inputs and body image representations (Cohen et al., 2009). Classical/normal cenesthetic 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 cenesthetic 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.

5. Uncited references

Cohen et al. (0000); Dimitri et al. (0000); Loby et al. (0000); Spinney (2007).

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References