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Effect of Intrathecal Baclofen Therapy Compared with DBS in Patients with Dystonic Cerebral palsy

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ABSTRACT

Dystonia is a symptom defined by involuntary and irregular contractions of the muscles, which cause movement disorders and postural problems. It is also common for these patients to have later changes in the musculoskeletal system and pain due to spasticity, dyskinesia, and dystonia. However, there remains no permanently effective treatment for patients with severe dystonia. Deep brain stimulation(DBS) in globus pallidus interna(GPi) is a good option for controlling dystonia. Intrathecal baclofen(ITB) has been known to reduce spasticity which did not respond to oral medications and botulinum toxin treatment. Many studies of ITB treatment have reported significant decreases in dystonia. We report a case that showed remarkable improvement in terms of the Short form 36 Health Survey version 2 (SF36), dystonia rating scale (DRS), modified Barthel index (MBI) and Visual analog scale (VAS) for pain after ITB pump (ITBP) implantation compared with DBS in patients with dystonic CP.

CASE REPORT

A 35-year-old female patient came to the rehabilitation clinics at our hospital with posterior neck pain and right upper extremity pain. She was experiencing severe dystonia in her neck and bilateral upper extremities; the DRS showed 66 on the dystonia movement scale, and 19 on the disability scale. In terms of spasticity, right upper extremity scored grade 2, left upper extremity scored 1+ and both lower extremities scored grade 1 on the MAS. She had 53 points on the MBI. She also suffered from pain in the posterior neck pain and both shoulder pain with a VAS of 8 scores. Although she has been treated with oral medication, it was no improvement for pain. After DBS was performed on the GPi, therapeutic outcomes were evaluated through DRS, MBI, SF36 and VAS. On the dystonia movement scale of DBS, the patient scored 66 before DBS, but 3,6 and 12 months later, she scored 72 which was worse than pre-DBS state. On the disability scale of DBS, the patient scored 19 before DBS, 27 one year after DBS and 22 six years after DBS (Table 1). MBI gradually decreased and it show persistent deterioration of functional level after DBS (Table 2). Spasticity was increased than pre-DBS state. The ITBP was operated for this patient who showed definitive positive response to the ITB test trials, and no adverse events. After the ITBP, starting with 50 μg, dose gradually increased to 130 μg. On the dystonia movement scale, the patient scored 72 before ITBP, but 3 months later, she scored 63. Two year later, the patient scored 64, which was no aggravation of dystonia. On the disability scale, the patient scored 22 before ITBP, but two year later, she scored 20 (Table 1). ADL was also improved in the MBI after the ITBP

(Table 2). Although she had severe posterior neck and shoulder pain scored VAS 8 before the ITBP, the pain score decreased to VAS 1 after the ITBP (Table 3). Post-ITBP satisfaction showed high scores when compared to post-DBS in all the items of SF36 (Table 3)

Table 1. Changes in dystonia rating scale after deep brain stimulation and intrathecal baclofen pump implantation

Dystonia rating scale	Pre-DBS	Post-DBS					ITB test trial		Post-ITBP			
		3 mo	6 mo	12 mo	24 mo	72 mo	Bolus inj.	Infusion	3 mo	6 mo	12 mo	24 mo
Dystonia movement scale	66	72	72	72	70	75	64.5	64.5	63	63	65	64
Disability scale	19	30	26	27	25	22	23	23	21	21	19	20

Mo, Months; DBS, Deep brain stimulation; ITB, Intrathecal baclofen; ITBP, Intrathecal baclofen pump implantation.

Table 2. Changes in modified Barthel index after deep brain stimulation and intrathecal baclofen pump implantation

	Pre-DBS	Post-DBS		ITB test trial	Post-ITBP		
		6 months	72 months		6 months	12 months	24 months
MBI	53	34	27	55	54	51	51

MBI, Modified barthel index; DBS, Deep brain stimulation; ITB, Intrathecal baclofen; ITBP, Intrathecal baclofen pump implantation.

Table 3. Comparison of the patient's response to the deep brain stimulation and Intrathecal baclofen pump implantation

	Post-DBS	Post-ITBP
Pain (VAS)	8	1
SF36		
Physical functioning	0	50
Role limitations due to physical health	0	75
Bodily pain	22.5	67.5
General health	60	70
Vitality	12.5	62.5
Social functioning	25	100
Role limitations due to emotional problems	0	100
Emotional well being	15	75
Physical component score	20.63	65.63
Mental component score	13.13	84.38

VAS, Visual analog scale; SF36, Short form 36 Health Survey version 2; DBS, Deep brain stimulation; ITBP, Intrathecal baclofen pump implantation.