

Deep Brain Stimulation Surgery of Amygdala in Autism – A Case Report

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Abstract

Autism, or autism spectrum disorder (ASD), refers to a broad range of conditions characterized by challenges with social skills, repetitive behaviors, speech and nonverbal communication. In patients with autism about 40 to 50% develop severe aggressive behavior. Pharmacological treatment is good for this but most patients do not benefit. In such patients Deep Brain Stimulation (DBS) surgery is a good option. This is the case report of one such case.

Introduction

Autism spectrum disorder (ASD) is characterized by highly variable cognitive ability and adaptive function, as well as diverse comorbid

behavioral symptoms that can be impairing. Aggression is a particularly significant challenge to caregivers and clinicians and could be seen in 35–50% [1]. This also causes problems with community engagement, relationships and independence. Strong predictors of aggression in typically developing (TD) individuals from longitudinal data include harsh parenting practices, low parent education, low IQ and male sex [2].

Case Report

In this report is a 28-year-old male patient diagnosed with autism spectrum disorder with associated low IQ and attention deficit disorder. He had problems with multiple social issues and had a score of 163 in the Indian Scale for assessment of autism (ISAA) (see table 1).

Parameter	Preoperative value	Postoperative value
Social Relationship and Reciprocity	39/45	19/45
Emotional Responsiveness	21/25	15/25
Speech	27/45	30/45
Behavior patterns	28/35	08/35
Sensory Aspects	19/30	17/30
Cognitive component	19/20	18/20
Total	163/200	97/200

Table 1.

The Indian Scale for Assessment of Autism (ISAA) mild AD: 70-107, moderate AD: 108-153, severe AD: Above 153.

The preoperative CARS score was 49/60 and the postoperative score was 27/60. The Global Assessment of Functioning, (GAF) scale preoperatively was 20 – 11. And the Postoperative score was 60 -51.

The Modified Overt aggression score also was calculated (Table 2).

Category	Sum score		Weights	Weighted Sum	
	Pre-oper	After 6/12		Pre-oper	After 6/12
Verbal Aggression	2	1	× 1	2	1
Aggression against property	3	0	× 2	6	0
Auto-aggression	2	0	× 3	6	0
Physical aggression	3	0	× 4	12	0
Total Score	10	1		26	1

Table 2.

Modified Overt aggression score (MOAS)

Psychometry showed attention span of less than a minute and moderate mental retardation preoperatively. Developmental age of the patient was about 4 yrs. The Autism Diagnostic Observation Schedule Generic (ADOS-G) pattern in this patient was also showing poor scores (Table 3).

Module	Pre-op score	After 6 months
1	11/18	11/18
2	18/28	15/28
3	19/26	11/26
4	21/26	17/26

Table 3.

Autism Diagnostic Observation Schedule Generic (ADOS-G)

The patient had been on various medical management for the last many years but condition has been deteriorating. He was on various medications including Risperidone, Aripiprazole, Clozapine, clonazepam and lorazepam, haloperidol, methylphenidate, and escitalopram in high doses but no benefit. So finally he was planned for deep Brain Stimulation (DBS) Surgery. He underwent this surgery in 2020. The target chosen was the basolateral nucleus of the amygdala. The post-operative period was uneventful.

Assessments after six months showed significant improvement in his aggression and also may autism features. His mother who is a doctor reported much improvement in speech and responsiveness and significant change in his aggressive behaviour (Table 1, 2, 3). At about one year the therapist also reported increased attention span up to 14 minutes (preoperatively it was less than a minute).

DISCUSSION

Autism is a neurodevelopmental disorder characterized by impairments in reciprocal social interaction, deficits in verbal and nonverbal communication, and a restricted repertoire of activities or interests. Studies have shown that children with autism had larger right and left amygdala volumes than control children [3]. Also studies also have shown that even though there were not much difference in the overall volume of the amygdala and its sub nuclei, there are significantly fewer neurons in the autistic amygdala overall and in its lateral nucleus.

In conjunction with the findings from previous magnetic resonance imaging studies, the autistic amygdala appears to undergo an abnormal pattern of postnatal development that includes early enlargement and ultimately a reduced number of neurons [4].

Anatomically the amygdala is a medial temporal structure and contains 13 nuclei that are grouped into three functionally different divisions of nuclei:

1. The basolateral group
2. The central group
3. The corticomedial group.

The basolateral nucleus (BLA) is a cortical-like structure in the dorsal amygdala, and it regulates behavioural and physiological responses to stress. The central amygdala (CeA) plays a crucial role in physiological responses to stressors, such as fearful stimuli, stressful stimuli, and some drug-related stimuli [5].

Three case-reports of DBS in four patients with treatment-refractory autism spectrum disorder (ASD) have been published. Sturm *et al.* 2012 [6] were the first to apply DBS to a patient with Kanner's autism and life-threatening self-injurious behaviour (SIB). The electrodes were placed in the basolateral nucleus of the amygdala (BLN). After 24 months, SIB and core symptoms of autism spectrum in emotional, social, and cognitive domains were improved, Segar, Chodakiewitz, Torabi and Cosgrove [7] reported beneficial effects of BLN DBS for a 24-year old patient with severe autism and Kleefstra syndrome, a rare genetic condition with TS and OCD-like symptoms. Three years after placement of the DBS devices, the patient continued to show improvements in her coprolalia, the involuntary and uncontrollable use of obscene language, social behaviour, and also her TS and OCD-like symptoms.

Stocco examined the effects of DBS in two patients with severe stereotypies. The first patient was a 19-year old woman whom received Gpi DBS. At 13 months post-DBS, her Hopkins motor stereotypy rating scale (JHMRS) decreased by 91.3% (from 46 to 4). The second patient, an 18-year old man, showed an initial decrease of his JHMRS score from 67 to 19 at 3-months follow-up after DBS of both the ALIC and GPi;

however, at 6-months post-surgery, his stereotypies gradually returned to baseline.

Two recent reviews also supported the role of DBS in aggressive behaviour in autism. In the first one Sinha, McGovern and Sheth [9] highlighted the microstructural, macrostructural, and functional abnormalities that emerge during brain development, which create a pattern of dysfunctional neural networks involved in socioemotional processing, how these findings implicate the BLA as a key node in the pathophysiology of autism and how BLA DBS for treatment of severe autism could be effective. Graat, Fige and Denys [10] opined that DBS of the amygdala might be more valuable for ASD patients with predominantly social dysfunctioning and SIB, whereas ASD patients with compulsive behaviours or stereotypies might benefit more from stimulation of striatal areas.

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