

CASE STUDY

Reduction in Obsessive Compulsive Disorder and Self-Injurious Behavior with *Saccharomyces boulardii* in a Child with Autism: A Case Report

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Abstract

This case report describes the effective use of *Saccharomyces boulardii* in a boy with autism spectrum disorder, obsessive compulsive disorder (OCD), and self-injurious behavior (SIB). Gastrointestinal dysfunction and OCD are frequent comorbidities in autism, which may share a common etiology resulting from a disturbance in normal gut microbiota. Alterations

in microbial diversity influence neuroinflammation and are linked to mood disorders, abdominal pain, and SIB. *S. boulardii* is a nonpathogenic probiotic yeast that supports a healthy microbiome, enhances immune function, and reduces diarrhea. Treatment with *S. boulardii* successfully reduced OCD and SIB symptoms in this child.

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Introduction

Autism spectrum disorder (ASD) is a neurodevelopmental disorder characterized by developmental impairments in multiple domains, which may include language, social communication, repetitive or rigid behaviors, cognitive function, and sensory sensitivity. Gastrointestinal dysfunction is a common comorbidity in ASD and is often presented as chronic diarrhea, constipation, and/or abdominal pain.¹ Children with ASD have imbalances in normal gut microbiota,² which are theorized to affect neurologic function via the gut-brain axis.³ Heightened pain sensitivity has been identified in persons with ASD, and children with autism with self-injurious behavior (SIB) exhibited significant increases in pain on a nonverbal pain rating scale.⁴ Increased microglial activation and elevated levels of inflammatory cytokines underscore the role of neuroinflammation in ASD⁵ and the relation to gut dysfunction. Prevalence of obsessive compulsive disorder (OCD) in ASD has been estimated at 2.6% to 37.2%.⁶

Neuroinflammation and alterations in gut microbiota are also associated with OCD.⁷

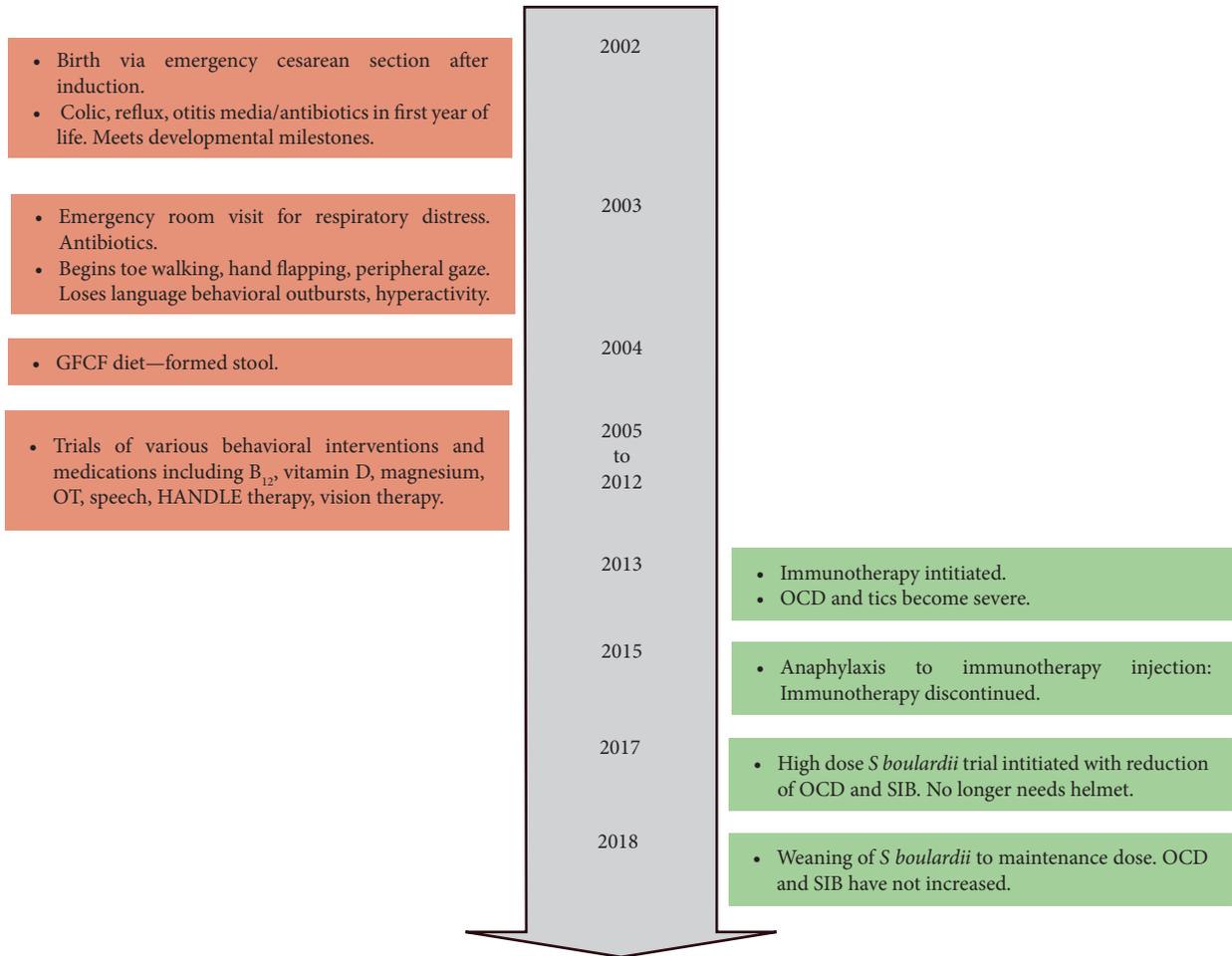
Saccharomyces boulardii is a safe and effective agent for the prevention and treatment of gastrointestinal diseases and is used most commonly to treat antibiotic associated diarrhea, *Clostridium difficile* infection, and irritable bowel disease. *S. boulardii* reduces pathogenic adhesion and colonization of the mucosa, acts as a prebiotic and probiotic to support growth of beneficial microbial species, modulates immune responses, and stabilizes gastrointestinal barrier function.⁸ This case reports on the successful utilization of *S. boulardii* to ameliorate severe OCD and SIB in a child with autism.

Case Presentation

A 16-year-old white male with a diagnosis of ASD was seen for increasingly severe OCD and SIB. Birth history is significant for induction and delivery by Cesarean-section with Apgars 9/9. As an infant, the child was reported to have colic, chronic diarrhea, frequent rashes, and otitis media (OM) treated with antibiotics. Developmental milestones were met until 15 months of age. Shortly after, he was admitted to the Emergency Department of the local hospital with respiratory distress after taking amoxicillin for OM. At 18 months of age, he began to regress, losing previously acquired language, toe walking, hand flapping, and exhibiting behavioral outbursts and hyperactivity. A gluten-free and casein-free diet was initiated at 26 months, which resulted in his first formed stool, and improved eye contact. He was formally diagnosed with ASD at 2.5 years of age. Bowel movements remained erratic. He had multiple allergies. At age 5 years, he began treatment at the clinic

Timeline.

Chief Complaint: 16-year-old-boy With Obsessive Compulsive Disorder and Self-Injurious Behavior Treated With *Saccharomyces boulardii*



Abbreviations: GFCF, gluten-free casein-free diet; OCD, obsessive-compulsive disorder; SIB, self-injurious behavior.

where he currently receives care. OCD, tics, and SIB became severe at age 11 years, at which time he began to wear a helmet to protect his head from constant head banging. Immunotherapy for allergies was initiated at age 11 years but was discontinued due to an anaphylactic response. The patient received supplements directed at gut and immune health including probiotics, vitamin D titrated to levels of 60 to 80 ng/mL, curcumin 600 mg BID, helminthic therapy, and bethanechol titrated for good gut motility. Additional treatment for documented and undocumented but clinically symptomatic yeast infections included fluconazole, itraconazole, nystatin, grape seed extract, and dietary restriction of simple carbohydrates, sugars, and fruits. He received a trial of inositol 4 to 12 grams/day for OCD behaviors. Magnesium for anxiety and neurologic stability was titrated between magnesium citrate and gastrointestinal neutral forms based on bowel movements. He also received trials of lorazepam PRN and clonazepam daily in the past

and was taking sertraline 75 mg QD at the time. Prior interventions included valacyclovir, cod liver oil, and methylcobalamin shots. Despite all of these interventions, OCD and SIB remained severe enough that he was often homebound. Laboratory evaluation was positive for elevated eosinophil counts and immunoglobulin E 4300 UI/m: (Centra lab, reference range 5 to 127 IU/mL), consistent with allergies and immune dysregulation. At age 15 years, a trial of *S. boulardii* was initiated because it has the unique benefits of promoting normalization of gut flora, improving immunomodulation, and managing recurrent diarrhea. *S. Boulardii* (Klaire Labs, 3 billion CFU per capsule) was initiated at 6 capsules daily: 2 at breakfast, 2 at lunch, 1 at dinner, and 1 at bedtime. Initial response was an exacerbation of symptoms attributed to Jarisch-Herxheimer reaction and treated with activated charcoal (Bulletproof) 1 capsule QID. After 1 week, *S. boulardii* was increased to 2 capsules QID, and the patient was able to discontinue use of the helmet for

2 days. He continued weekly increases of *S boulardii*, reaching a final dose of 12 capsules BID equivalent to 72 billion CFU daily. With each increase there was a transient uptick in OCD and SIB, which was addressed with activated charcoal. During this time, use of the helmet diminished markedly and OCD symptoms decreased to a parent rating of 2/10. After 3 months at 24 capsules of *S boulardii*, the patient began weaning down by 4 capsules per week to a dose of 3 capsules BID. SIB occurs once or twice per week for approximately 10 minutes each, whereas prior to the *S boulardii* treatment, SIB occurred multiple times per day and would last for more than 1 hour each time. He has maintained all gains to date.

Discussion

Although the exact etiology of ASD had yet to be elucidated, it is recognized that both genetic and environmental influences play a role, and that perturbations of the bidirectional gut-brain axis are an important area of exploration. GI dysfunction, frequently presenting as diarrhea, constipation, or abdominal pain has been identified in up to 70% of children with autism with increased symptoms correlated with ASD severity.⁹ Increased pain is correlated with SIB. Both mouse and human models of autism identify alterations in the normal composition of gut microbiota in ASD. Antibiotic use disrupts the normal microbial balance, and decreased diversity in gut flora has been associated with C-section birth.¹⁰

Gut microbiota have immunoregulatory effects on the production of inflammatory cytokines such as tumor necrosis factor α , immunoglobulin 6, and immunoglobulin 1. Elevations in these cytokines are linked to mood disorders such as anxiety and depression. Research regarding the role of imbalanced gut ecology and related immune activation in OCD provides parallels to autism and may be a factor in the high frequency of comorbidity. The immunological alterations in OCD that reduce serotonin (5-HT) synthesis via activation of indolamine-2, 3-dehydrogenase (IDO) have been described by Marzetti et al.¹¹ Increased IDO will shunt tryptophan to kynurenic and quinolinic acids which have excitotoxic properties. An abrupt onset of OCD is a hallmark of pediatric acute-onset neuropsychiatric syndrome (PANS), a disease in which neuroinflammation is suspected to play a role in the majority of cases.¹² Fungal gut pathogens are also implicated in mental health disorders. Levels of *Candida albicans* IgG were significantly elevated in male patients with schizophrenia and bipolar disorder as compared with controls.¹³

S boulardii is a nonpathogenic, transient yeast that increases the activities of intestinal brush border enzymes, enhances stool concentrations of short-chain fatty acids that nourish colonic mucosal cells, raises intestinal secretion of secretory IgA and immunoglobulin receptors, and reduces diarrhea. It functions as both a prebiotic and probiotic and has the ability to reduce adhesion of pathogenic species such as *C albicans* in the intestinal

tract. *S boulardii* survives gastric acidity and exposure to antibiotics. It safe for use in adult and pediatric populations, however, some instances of fungemia has been documented in severely immunocompromised patients.¹⁴

Conclusion

There are currently no widely recognized effective treatments for the often debilitating behavioral and gastrointestinal manifestations of ASDs. *S boulardii* offers great promise due to its ability to potentiate desirable microbial balance in the gut, positively modulate immune response, and control chronic diarrhea, while maintaining a strong safety profile. More studies of *S Boulardii* in autism and OCD are warranted.

Acknowledgements

Victoria Kobliner, MS, RDN, conducted the literature review and was the primary contributor to the creation of the manuscript. Elizabeth Mumper, MD, conducted the chart review, provided feedback, revisions, and editing of the manuscript. Sidney M. Baker, MD, conducted chart review and provided feedback, revisions, and editing of the manuscript. Victoria Kobliner, MS, RDN, is the owner of Holcare Nutrition, a private practice specializing in functional nutrition therapy. It is located at 3 Hollyhock Road, Wilton, CT, USA 06897. Elizabeth Mumper MD, FAAP, IFMCP, is a pediatrician and chief executive officer of Advocates for Children, Advocates for Families, and the Rimland Center for Integrative Medicine at 2919 Confederate Avenue, Lynchburg, VA, USA 24501. Sidney M Baker, MD, is located at 71 Ferry Road, Sag Harbor, NY, USA 11963. The authors would like to acknowledge the patient and his family for their willingness to share their story.

Author Disclosure Statement

Written informed consent was obtained from the parent of the minor child for publication of this case report. The authors declare no competing interest/financial disclosures.

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