

Case Report



Integrated approach in the postoperative care of Currarino Syndrome: A case report

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ABSTRACT :

Background- Currarino syndrome is a congenital disorder characterized by a triad of features: a presacral mass, anorectal malformation, and a sacral bone defect. This case report discusses the integrated Ayurvedic and modern management of a pediatric patient with persistent postsurgical constipation due to Currarino syndrome. In cases of congenital anorectal stenosis, Currarino triad should be considered as a potential underlying diagnosis and evaluated through thorough physical examination and appropriate imaging. Long term management involves addressing defecation and urinary problems which may be influenced by the degrees of sacral agenesis. **Materials and methods-** A 9-year-old male presented with irregular bowel habits since birth. He had a history of posterior sagittal anorectoplasty. X-ray of lumbosacral spine showed defect from third sacral vertebra. According to modern science and traditional *Ayurvedic* medicine, the case was identified as constipation and *Vibandha*, respectively. The patient received an integrated treatment plan of Proctoclysis enema and *Matra Basti*. **Results-** The symptoms were relieved completely after 1 month of treatment with no adverse effects. **Conclusion -** *Vibandha* was managed successfully with the integrated approach of Proctoclysis enema and *Matrabasti* along with ayurvedic oral medication for the period of 1 month with no recurrence after 2 years of follow-up.

KEYWORDS: *Vibandha*, Currarino syndrome, *Matra Basti*, Case Report

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1. INTRODUCTION

Currarino syndrome is a rare genetic disorder typically involving a combination of anorectal malformations, sacral bony defects, and a pre-sacral mass, most often associated with mutations in the HLXB9 gene located on chromosome 7q36 (Currarino et al., 1981). [1] Children are commonly affected; with females generally being affected more frequently than males. [2] Anorectal malformations are defined by the relationship of the rectum to this complex and include varying degrees of stenosis to complete atresia. Although anorectal malformations are relatively common in pediatric populations, their surgical correction continues to pose challenges due to the variability in anatomical presentations and associated complications. [3] It is important to consider the importance of the sphincter complex, in understanding the spectrum of anorectal anomalies. This complex consists of the puborectalis, levator ani, external and internal sphincters, and the superficial external sphincter muscles, all converging at the rectum (Kukreja, 2013, pp. 276–277). [4]

During the last several decades, significant advances have been made in the understanding and treatment as well as in the correction of anorectal malformations. Despite these advances, the primary goal of the surgeon remains a very evasive one. The main aim of the surgical treatment for these patients is to achieve a repair and to create a reconstructed anatomy which provides a nearly normal anal outlet for the patient, in an attempt to provide these children with a good quality of life. [5]

The majority of patients who undergo surgical repair for an anorectal malformation experience some level of functional defecation disorder. [6]

1. Intact sensory mechanism lacking in many cases
2. Abnormal voluntary muscle structures leading to incontinence and improper bowel habits

Here we report a case of post-operative complication of Currarino syndrome. This case report is “Unique in demonstrating the successful long-term management of complex post-surgical constipation associated with Currarino Syndrome through an integrated multidisciplinary approach, offering valuable insights into combining modern medicine and traditional Ayurvedic principles for such challenging conditions”.

2. CASE REPORT

A 9-year-old male child, diagnosed with Post-operative case of Currarino syndrome was treated at SDM College of Ayurveda and Hospital, Hassan with Hospital ID-IP-063057 from 15th December 2022 to 15th January 2023. The patient presented with the complaints of incontinency of bowel since 5 years and incomplete evacuation of stools since birth. Associated complaints were pain abdomen and distention of abdomen. There was no significant family history and genetic history; he was not actively participating in daily activities as he was not able to perform his routine activities.

Past history of altered and difficulty in passing stools and was treated with laxatives on and off for first 2 years of life. Then with the help of detailed radiological investigations patient was diagnosed with Currarino syndrome.

Previous radiological investigation (MRI) was suggestive of S1 S2 and S3 vertebrae are visualized. Rest lower sacral vertebrae and coccyx are not visualized, suggestive of partial sacral agenesis. Large bony defect is seen in anterior sacral vertebrae (S2 S3) Anterior sacral vertebral defect measure 0.8 to 0.9cm. Large localized fluid collection /CSF collection is seen in pre-sacral space. Localized fluid /CSF space in pre-sacral is measuring 8.5 X 3.4 X 4.0 cm (CC x AP x RL). Pre-sacral collection is causing anterior and right lateral displacement of rectum. Antero superior displacement of urinary bladder was noted.

Past treatment history suggested that patient underwent for Posterior sagittal anorectoplasty with excision of pre-sacral mass under general anesthesia followed by manual anal dilatation at private hospital in 2015.

Operative finding were as follows: Anal stenosis 1.5" in length. Large pre-sacral mass 5X5cm lobulated, with putty like material- Dermoid. Sacral anomaly-Absent coccyx and some pieces of sacral bone. Neoanus created with 13 number hegars calibration.



Figure 1 – Showing the Anal canal stenosis

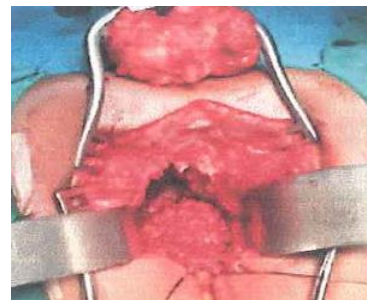


Figure 2 – Showing the Excised pre-sacral mass during operating time

During post-operative days watery discharge from drain persisted for 14 days and there no features of fever and vomiting. Pre-sacral collection and biopsy proved dermoid cyst and meningocele like sac proving the case to be of Currarino triad.



Figure 3 – Showing Neo-anus created in Post-operative period

Five years after surgery patient came with complaints of altered bowel habit and constipation and on detailed clinical examination patient was diagnosed with post-surgical anal stricture.

Clinical findings-

The general examination indicated that the patient was moderately built and nourished. Systemic examination showed that the patient was conscious and well-oriented to time and place and was actively participating in day-to-day activities. Vital signs revealed a blood pressure 100/60 mmHg, a pulse rate of 96 beats

per minute, a respiratory rate of 20 cycles per minute, and a body temperature of 98.2°F.

The local examination revealed that on inspection of anal region-Previous surgical scar was seen at natal cleft and anal verge. On digital rectal examination hyper tonicity was noticed and proctoscopy was not carried out due to hyper tonicity.



Figure 4 – Showing deformity in the anal verge along with post-operative scar at 6 O’clock position

After overviewing the symptoms, the present case was diagnosed with *Sanniruddha Guda* as per *Ayurveda* and Currarino syndrome as per modern. Overall prognosis of the disease when associated with congenital anomaly of sacral bone is difficult to treat with expected complications and high recurrence rate.

3. INTERVENTION

The treatment plan developed using a integrated approach, combining modern medicine and Ayurveda. The prognosis, complications and risks to the patient were explained to the patient’s parents and treatment was initiated after obtaining their consent and investigations.

Table 1: Showing Therapeutic Intervention and Oral Medicine

SI.NO.	Plan Of Care	Procedure	15/12/2022 to 17/12/2022	18/12/2022 to 25/12/2022	26/12/2022 to 15/01/2023
1.	Modern Medicine	Proctoclysis enema 50 ml in the morning for 3 days	✓		
2.	<i>Avagaha Sweda</i> with luke warm water followed by Kegels exercise BD		✓	✓	✓
3.	<i>Matra Basti</i> with <i>Yashtimadhu Taila</i> 50 ml in morning			✓	
4.	Tab <i>Kamaduga Mukta</i> 250mg BD Before food		✓	✓	✓
5.	<i>Haritaki Khanda</i> 10gm HS with Warm water before food		✓	✓	✓

Timeline:

The detail of progress of the disease along with treatment is provided

Table 2: Disease progression and treatment

SI.NO.	Year	Symptoms	Intervention
1.	Jan 2014 to Mar 2014	Pain and difficulty during defecation	On laxatives for 3 months
2.	09 Oct 2015	Constipation	Posterior sagittal anorectoplasty with excision of

		Diagnosed with Anal stenosis	pre-sacral mass from
3.	Apr 2016 to May 2016	Symptoms persists	Manual anal dilatation and laxatives
4.	18 Dec 2022 to 25 Dec 2022	Symptoms persists	<i>Avagaha Sweda</i> followed by <i>Matrabasti</i> with <i>Yashtimadhu Taila</i> for 8 days
5.	Sept 2023	No recurrence of the symptoms	Improved quality of life
6.	Nov 2024	No recurrence of the symptoms	Improved quality of life

Follow Up and Outcomes

The Patient was called regularly for the follow ups to assess reduction in the symptoms and quality of life. During the follow up period of 30 days patient was not complaining any of the symptoms. There was improved quality of life was noticed. The patient was called after 6th month, 12th month, 18th month to check for reoccurrence and there was no reoccurrence of the symptoms seen.

4. DISCUSSION

The Currarino Triad was first described in 1981 by pediatric radiologist Guido Currarino and consists of sacrococcygeal defect, pre-sacral mass and anorectal malformation (Caro-Domínguez, et al., 2017). [7] The true incidence of this triad is unknown, with fewer than 250 cases documented in the literature. Owing to phenotypic variability both within and between families, it is believed that many cases may go undetected (Vargas-González et al., 2008). [8] Clinically, Currarino Triad typically presents at birth with an imperforate anus or, in some cases, persistent constipation due to anorectal stenosis. Surgical management primarily aims to correct the anorectal malformation and remove any associated pre-sacral mass. However, several risks and postoperative complications must be considered,

including potential nerve injury that may lead to fecal incontinence, bladder dysfunction, anal stenosis, and other related issues.

In the present case report patient had suffered with constipation for long term which was one of the post surgical complication and patient had underwent several treatment modalities from the modern science but did not get relieved completely. To treat the same he has approached to Ayurveda and which was treated with Proctoclysis enema followed by *Avagaha Sweda* and *Matrabasti* was given. Proctoclysis enema helps to soften hardened feces and with less quantity it triggers defecation reflex by the presence of fluid in the rectum, which was used for cleansing the bowel and *Basti* was used to strengthen the rectum and anal canal which is one of the *Marma Sthana*. As stated in Charaka Samhita, *Basti* therapy is considered the most effective treatment for balancing aggravated *Vata Dosha*, especially in conditions affecting the lower gastrointestinal tract. [9]

Probable mode of action of *Avagaha Sweda*: *Avagaha Sweda* helps in vasodilatation of the local blood vessels leading improved circulation and along with this when drug will be administered through per rectal route helps in drug absorption and gives both local and systemic

effects. Kegels exercise strengthens the muscles that support your uterus, bowels, bladder and rectum.

Probable mode of action of *Basti*: The unctuous and warming properties of medicated oils used in *Matra Basti* therapy effectively counter the dry, light, and cold attributes of aggravated *Vata dosha*. This promotes bowel lubrication and regular defecation, aligning with the Ayurvedic principle of *Vatanulomana* (downward movement of *Vata*). [9]

Garbha, Vata, Mutra Nishkramana kriya is done by *Pakvashayagata Apana Vata*, if *Apana Vata* is normalized it does the *Vatanulomana* and relieves the constipation. Once the aggravated *Vata* is expelled, *Matra Basti* helps restore balance and promote overall health.

Adverse effects: No adverse or unintended effects were observed during the course of the study.

5. CONCLUSION

The current case involved a long-standing issue of constipation in a 9-year-old boy diagnosed with post-operative Currarino Syndrome. The intervention used an integrated approach, primarily featuring *Matra Basti*, which is a form of *Shamana Karma* in Ayurveda, along with Proctoclysis enema. The treatment lasted 1 month and resulted in complete symptom relief. The patient was monitored for 2 years with no return of symptoms and an improved quality of life. No negative effects were seen during or after the treatment.

Key Findings:

1. Successful long-term management of post-surgical chronic constipation in Currarino Syndrome using an integrated approach.

2. Efficacy of *Matra Basti* as a safe and effective Ayurvedic intervention for complex bowel disorders.

Improvement in quality of life for the patient.

Declaration of Patient Consent – The authors confirm that they have acquired a patient consent form, in which the patient or caregiver has granted permission for the publication of the case, including accompanying images and other clinical details, in the journal. The patient or caregiver acknowledges that their name and initials will not be disclosed, and sincere attempts will be undertaken to safeguard their identity. However, complete anonymity cannot be assured.

Patient perspective - The patient's parents were very satisfied with the combined treatment approach. They noted a clear improvement in the child's bowel habits and overall quality of life. They said that the long-standing issue of constipation, which caused a lot of distress, was finally resolved. They valued the teamwork of the healthcare providers and the gentle nature of the Ayurvedic treatment.

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