



Case Report

Ayurvedic management of chronic constipation in Hirschsprung disease – A case study

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ABSTRACT

Hirschsprung disease (HSCR) or congenital intestinal aganglionosis is characterized by complete absence of neuronal ganglion cells from a portion of the intestinal tract, most commonly in the large intestine. The main sign or symptom of HSCR is constipation usually appearing shortly after birth. This constipation is chronic in nature and usually not relieved with laxatives. The present case is of a patient having HSCR which was successfully managed with Ayurvedic treatment. A four year old boy with complaint of severe constipation, abdominal pain, abdominal distension and occasional vomiting was treated with *Panchakarma* procedures and Ayurvedic oral drugs. The Ayurvedic diagnosis of the case was *Pakvasayagata vata*. *Shashtikashali pinda swedana* (sudation with medicated cooked bolus of rice) and *Matra basti* (enema with medicated oil) with *Ashwagandha taila* (Ayurvedic medicated oil) was given for first 16 days. From the 2nd month of treatment, *Matra basti* was administered daily for 3 months in the dose of 25 ml. In 5th and 6th month *Matra basti* was administered on alternate days in the dose of 25 ml. From the 7th month *Matra basti* was administered once weekly in the dose of 25 ml. In 14th month *Shashtikashali pinda swedana* and *Erandmooladi yapna basti* (medicated enema) was given for 16 days. Eight scales based Medical outcome study (MOS) – 36 item short form - health surveys was periodically assessed for outcome which shows good improvement. Experience of this case showed that HSCR may satisfactory be managed with Ayurvedic treatment.

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1. Introduction

Hirschsprung disease (HSCR), or congenital intestinal aganglionosis, is a birth defect characterized by complete absence of neuronal ganglion cells from a portion of the intestinal tract most commonly large intestine [1]. In a child suffering from HSCR, stool moves normally up to the part lacking nerve cells, then in that portion, the stool moves slowly or becomes stagnant. The main sign or symptom of HSCR is constipation or intestinal obstruction, usually appearing shortly after birth. The affected infants frequently present this symptom in the first two months of life and the early symptoms of impaired intestinal motility such as failure to pass meconium within the first 48 h of life. The late symptoms are constipation, emesis, abdominal pain or distention, and occasionally diarrhea. Most often, an infant or a child with HSCR will also have other symptoms, including growth failure or unexplained fever.

The incidence of HSCR is approximately one in 5000 live births [2]. However, the incidence varies significantly among ethnic groups (1.5, 2.1, and 2.8 per 10,000 live births in Caucasians, African-Americans, and Asians, respectively) [3]. The cause of HSCR is most commonly attributed to defective craniocaudal migration of neuroblasts originating from the neural crest during the first twelve weeks of gestation, resulting in functional intestinal obstruction [4]. Genetic defects can increase the chance of a child developing HSCR. Children with Down syndrome and other medical problems, such as congenital heart defects, are at much greater risk. No testing is available that can diagnose a child while the mother is pregnant.

HSCR is a life-threatening illness, and treatment requires surgery. Generally, patients can manage this condition through the use of cathartic agents. However, at some point, the dilated proximal colonic segment may decompensate secondary to the distal obstruction and patients may experience rapidly worsening constipation or even acute obstruction. Here is a case report of a child who suffered from HSCR and was effectively managed with Ayurvedic medications and *Panchakarma* procedures.

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2. Patient information

A four year old boy with complaint of severe constipation, abdominal pain, abdominal distension and occasional vomiting visited in O.P.D. of department of *Panchakarma*, National Institute of Ayurveda, Jaipur. He had history of delayed passage of meconium at birth and was managed successfully with soap and water enema. Since then patient was not being able to pass stools without enemas. Patient's developmental milestones, chest and vital signs were normal but he had distended abdomen. The height of patient was 100.2 centimeters (cm) and the weight of patient was 14.5 kilograms (kg). His appetite was normal. Patient had normal micturition. The patient had undergone for consultations in All India Institute of Medical Sciences (AIIMS), New Delhi two months before, where he was diagnosed as a case of HSCR and surgical management was recommended. He was only child of his parents and none of his parent was affected with these symptoms.

2.1. Clinical findings

On physical examination, patient was anxious and his tongue was uncoated. Patient had *Vata-pitta prakriti* with *Avara samhanana* (lowermost body constitution), *Avara sara* (~lowermost purest body tissue), *Avara satmya* (lowermost homolagation), *Avara satva* (lowermost mental strength), *Madhyam vyayamshakti* (middlemost capability to carry on physical activities), *Madhyam aharshakti* and *Jaranshakti* (middlemost food intake and digestive power). The patient demonstrated normal gait. On neurological examination higher mental function and speech were normal. All cranial nerves were intact. On motor examination, bulk, tone, power and co-ordination of arms and legs were normal bilaterally. Joint position sense and vibration sensation was normal bilaterally. Upon abdominal examination, patient demonstrated marked abdominal distention with palpable dilated loops of colon. Rectal examination revealed an empty rectal vault and resulted in the forceful expulsion of fecal material upon completion of examination. Complete blood count (CBC), Thyroid profile and tissue transglutaminase-IGA (TTG) were within limits.

3. Timeline

A detail of the case study and follow up is given in [Table 1].

Table 1
Timeline.

Year	Incidence/intervention
2012	Patient had severe constipation since childhood.
2014–2015	Patient was consulted for chronic constipation in J.K. Loan hospital and medical college Jaipur and Imperial hospital and research center Jaipur. Patient was suspected to suffer from HSCR. Patient was advised conservative treatment.
February–March-2016	Patient was consulted in Pediatric surgery department of AIIMS New Delhi. Diagnosis of HSCR was confirmed in AIIMS New Delhi. Patient was advised to go for biopsy and colostomy.
April-2016	Patient was unwilling for surgery. Patient visited O.P.D. of National Institute of Ayurveda Jaipur for these problems and was advised for administration of <i>Panchakarma</i> Procedures.
12/04/2016–27/04/2016	<i>Shashtikashali pinda svedana</i> was done for 16 days along with <i>Matra Basti</i> for 16 days. Selected Ayurvedic oral drugs- <i>Vrihatavatachintamani ras</i> and <i>Drakshaveleha</i> twice a day were also prescribed along with these <i>Panchakarma</i> procedures. There was clinical improvement in patient condition after one month of therapy. Same oral medication was provided to the patient.
12/05/2016–11/08/2016	<i>Vrihatavatachintamani ras</i> was discontinued from 12/07/2016. <i>Matra basti</i> with <i>Ashwagandha taila</i> once in a day was continued for three months.
12/08/2016–11/10/2016	<i>Drakshaveleha</i> twice a day was continued as oral medicine. <i>Matra basti</i> with <i>Ashwagandha taila</i> was given on alternate days for two months.
12/10/2016–11/05/2017	<i>Drakshaveleha</i> twice a day was continued as oral medicine. <i>Matra basti</i> with <i>Ashwagandha taila</i> was given once a week.
14/12/2016	Serum bilirubin both direct and indirect, serum glutamic oxaloacetic transaminase (SGOT), Serum glutamic pyruvic transaminase (SGPT), serum creatinine ESR,CBC, serum albumin, serum calcium, serum electrolytes, serum phosphorus and total lipid profile were investigated. These were within limit. Barium enema study demonstrates delayed emptying time and a funnel-like transition zone between proximal dilated and distal constricted bowel –suggestive of Hirschsprung disease.
17/05/2017	Above hematological and biochemical parameters were reinvestigated which were within limit.
12/05/2017–28/05/2017	<i>Shashtikashali pinda svedana</i> was done for 16 days along with <i>Erandmooladi yapana basti</i> for 16 days. <i>Drakshaveleha</i> twice a day was continued as oral medicine.

4. Diagnostic focus and assessment

The patient was a known case of HSCR. It was confirmed by previously done anorectal manometry and barium enema that show a dilated proximal colon with empty rectum and barium enema study that demonstrate delayed emptying time and a funnel-like transition zone between proximal dilated and distal constricted bowel (Figs. 1 and 2) [5]. History of delayed passage of meconium at birth and empty rectal vault and forceful expulsion of fecal material just after completion of rectal examination in the case were suggestive of HSCR. *Pakvasayagata vata* was considered as Ayurvedic diagnosis which is included in *Vatavyadhi* [~neurological, rheumatic and musculoskeletal diseases]. *Antrakoojana* (bowel sounds), abdominal pain, *Atoop* (flatulence), difficulties in passing urine and stool, *Anaaha* (~abdominal distension) and *Trikvedana* (pain around sacral region) are the symptoms of *Pakvasayagata vata*. [6] Acute mega colon, chronic mega colon, constipation, hypothyroidism, intestinal motility disorders, celiac disease, irritable bowel syndrome and toxic mega colon were the differential diagnosis for the case [7]. Thyroid profile and tissue transglutaminase-IGA (TTG) were within normal limit which excluded the diagnosis of hypothyroidism and celiac disease.

5. Therapeutic intervention

Line of treatment for *Pakvasayagata vata* is similar to general line of management of *Udavarta* (~abdominal distention due to constipation and other causes). *Abhyanga* (massage) with *Shita jwarokta* oil (oil used for *Shita jwara*), *Svedana* (sudation), *Varti* (suppository), *Niruha basti*, *Snehana*, *Virechana* (mild purgation), use of *Anulomak* (carminative) foods are indicated for the treatment of *Udavarta* [8]. Since the patient was in his childhood, mild form of different *Panchakarma* procedures was used. Patient was treated with *Mridu abhyanga* (mild massage) and *Mridu svedana* (mild sudation) and *Matra basti*.

Abhyanga (massage) with *Chandanabalalakashadi Taila* and *Mridu svedana* with *Shashtikashali pinda svedana* were done for 16 days. *Matra basti* with *Ashwagandha taila* were also prescribed for 16 days [Table 2]. After completion of these *Panchakarma* procedures a rest of 14 days from all these procedures was given to the patient. From the 2nd month of treatment, *Matra basti*

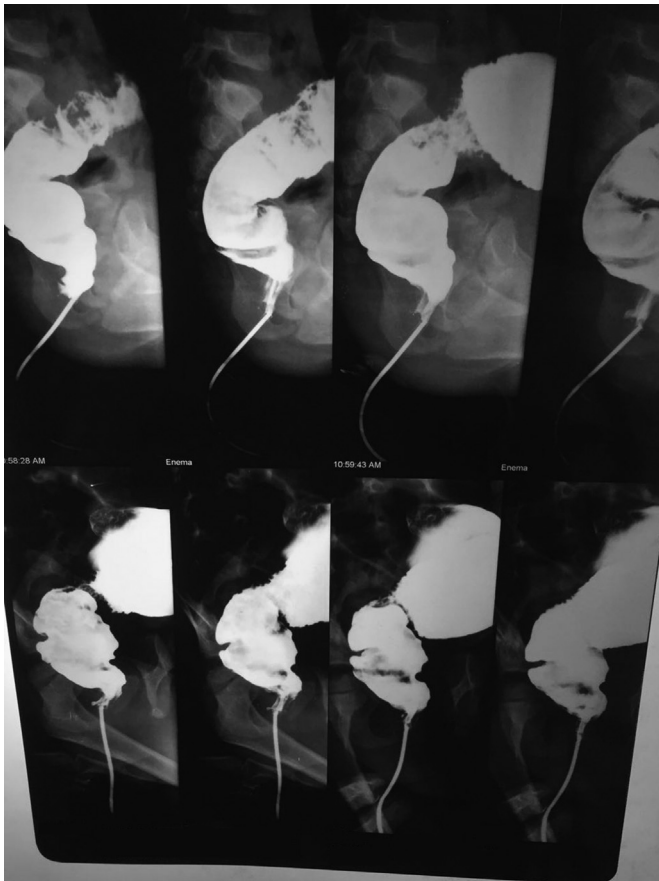


Fig. 1. Barium enema study.

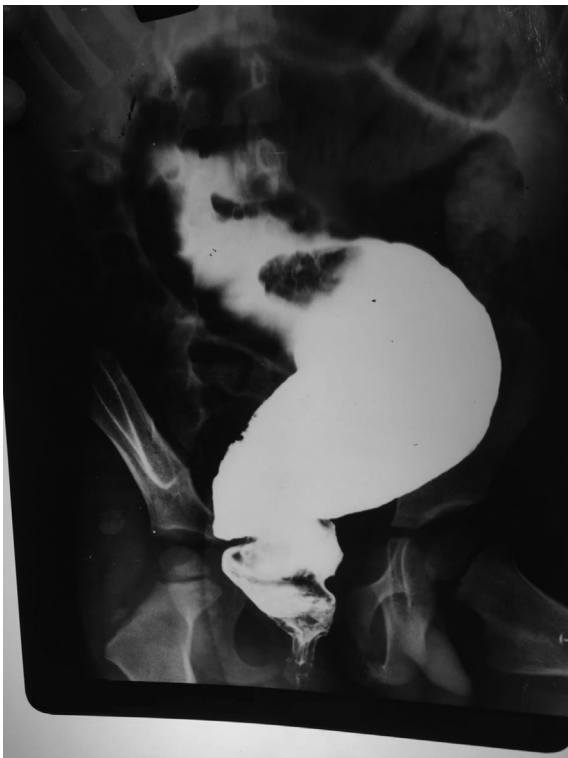


Fig. 2. Funnel like transition zone between proximal dilated and distal constricted bowel.

was administered daily for 3 months in the dose of 25 ml. In 5th and 6th month *Matra basti* was administered on alternate days in the dose of 25 ml. From the 7th month *Matra basti* was administered once weekly in the dose of 25 ml. *Mridu abhyanga* with *Chandanasbalalakshadi taila* and *Mridu svedana* with warm cloth (*Patta svedana*) were done as *Poorvakarma* (preoperative procedure) of *Matra basti* administration. After completion of one year of treatment, *Shashtikashali pinda svedana* was done for 16 days along with *Erandmooladi yapana basti* for 16 days. After completion of these *Panchakarma* schedule patient was advised to take *Matra basti* with *Aswagandha. taila* once weekly. Along with these interventions Ayurvedic oral medications-*Vrihatavatachintamani ras* 50 mg, with honey twice a day and *Drakshaveleha*-5 g twice a day were given regularly from 1st day up to 3 months [Table 3]. Only from the 4th month, *Drakshaveleha*-5 g twice a day is continue to till date.

6. Follow up and outcomes

Patient condition was assessed on different intervals on parameters like – child global health and ease in defecation. Good relief in constipation was noted in the patient [Table 4]. The patient had regular defecation in toilet after first *Panchakarma* schedule. No painful and hard bowel movement was noticed. Medical outcome study – Short form 36 (MOS SF-36) survey items were assessed for outcome which shows improvement [9]. There was increase of 7.5 cm in height and 2.7 kg in weight of the patient during the course of treatment. No concurrent Allopathic medication was administered during this period. Outcome measure was assessed on baseline, after completion of first *Panchakarma* schedule and at the completion of 4th month, at the completion of 6th month, 9th month and one year. There was improvement in quality of life and in non-specific abdominal pain. There was ease in defecation and bowel movement was regular. Patient was able to defecate every day and there was no pain in defecation. Patient felt some abdominal distension when *Matra basti* was not administered weekly so it is regularly being administered weekly till date. He did not suffer from any concurrent diseases during the course of period. Serum bilirubin both direct and indirect, serum glutamic oxaloacetic transaminase (SGOT), Serum glutamic pyruvic transaminase (SGPT), and serum creatinine that were tested on December 14, 2016 for assessment of safety profile of treatment were within limits. ESR, CBC, serum albumin, serum calcium, serum electrolytes, serum phosphorus and total lipid profile were also within limits. These investigations and SGPT, SGOT were also within limits on May 17, 2017, however serum alkaline phosphatase was increased which is normal for growing children.

7. Discussion

HSCR is a congenital disorder with main symptoms of severe constipation. Several oral drugs are in use for relieving these symptoms as these drugs are convenient and conventional. However, unlike healthy children and infants, children with HSCR typically do not respond to constipation medicines given by mouth. Various soaps enema are in use for these types of children for temporary relieving constipation but these are painful to the patients and there is also the rebound constipation. Some surgical options are also available for the disease but more postoperative complications and lower quality of life are the major concern to adopt the surgical procedures. Hence, treatments based on Ayurvedic principles may be suitable for the disease. Similar conditions are mentioned in description of *Pakvasayagata vata* disease. The line of management of *Udavarta* was adopted to treat this case, as line of management of *Pakvasayagata vata* is similar to the

Table 2
Panchakarma procedures given to a case of HSCR.

Panchakarma procedures	Method of preparation	Method of application	Days of treatment
<i>Shastikashali Pinda Svedana</i>	300 g of <i>Shashtika shali</i> (specific variety of rice, which is yielded after 60 days) is cooked with 1.5 L of milk and decoction of <i>Bala moola</i> (root of <i>Sida Cordifolia</i>). This mixture is to be kept in four pieces of cloth to make 4 boluses. Another portion of milk and decoction of the same quantity should be mixed and heated in low temperature to dip the above boluses for warming.	Massage with <i>Asvagandha taila</i> was done on whole body for 15 min followed by whole body massage for 45 min with the help of a cotton bag filled with bolus of processed rice.	16 days
<i>Matra Basti</i>	<i>Saindhava</i> salt 1 g, <i>Satahva</i> (Anethum sowa Kurz) 5 g, <i>Ashwagandha Tail</i> 25 ml, This emulsion is heated gently in a water bath.	Given after meal with 6 no. catheter and 50 ml syringe. Before <i>basti</i> procedure <i>Mridu abhyanga</i> (mild massage) and <i>Svedana</i> (fomentation) with slight warm cotton pad.	<i>Matra basti</i> was given for first 16 days, Daily from 2nd months to 4th months, From 5th to 6th days on alternate days. From 7th month to 12th month once in a week. Total 16 <i>basti</i> was given daily. (From 12/05/2017-to 28/05/2017)
<i>Erandmooladi yapana basti</i>	<i>Saindhava</i> salt 5 g, honey 25 g, <i>Aswagandha tail</i> 25 ml, <i>Panchatikta Ghrita</i> 25 ml, <i>Kalka</i> (paste)-10 g and milk processed with <i>Erandmooladi yapana basti</i> , <i>Kwath</i> (decoction) drugs 200 ml. Powdered rock-salt is added to honey and stirred. Then oil and <i>Ghrita</i> are added to this mixture and again stirred. Then paste followed by decoction is to be added and mixed properly to make homogenous emulsion. This emulsion is heated gently in a water bath.	Given before meal with <i>basti yantra</i> .	

Table 3
Ayurvedic treatment given to a case of HSCR.

Name of the drug used orally	Composition	Dose	Anupana	Days of treatment
<i>Drakshaveleha</i> (procured from market, manufactured by Dabur, India company)	<i>Draksha, Jaiphala, Javitri, Ela, Vanslochan, Lavanga, Dalchini, Tejapatta, Nagakeshar, Kamalgatta</i>	5 g twice a day	milk	From 1st day to till date
<i>Vrihatavatachintamani ras</i> (procured from market, manufactured by Dabur, India company)	<i>Swarna, Raupya, Abhraka, Moti, Praval, Lauha, Parad, Gandhak</i>	50 mg twice a day	honey	From 1st day up to 3 months

Table 4
Scoring of eight scales for the case of HSCR based on medical outcome study -Short form 36 (MOS SF-36) survey.

Scale	Scores ^a dated 12/04/2016	Scores ^a dated 12/05/2016	Scores ^a dated 12/08/2016	Scores ^a dated 15/12/2016	Scores ^a dated 15/03/2017
1. Physical functioning	5.55	61.11	77.77	100	100
2. Role limitation due to physical health	90	00	00	00	00
3. Role limitation due to emotional problem	33.33	66.67	66.67	100	100
4. Energy/fatigue	7	48	66	100	100
5. Emotional wellbeing	22	42	62	100	100
6. Social functioning	12.5	30	70	100	100
7. Pain	20	80	100	100	100
8. General health	10	40	80	100	100

^a Scoring by the instrument-RAND 36-item Health Survey 1.0.

management of *Udavarta*. The primary goals of Ayurvedic treatment in the case was to maintain normal fluid and electrolyte balance, to minimize bowel distension, to relieve the constipation, to manage complications and to improve the quality of life. *Chandanabalalakashadi taila* is having the property of *Saptadhatu vivardhana* (nourishment to body tissue) and *Kasa Swas kshaya haram* (effective in cough, dyspnea and emaciation). It is effective in *Pitta kaphaja* diseases, in *Vataja* disorders of emaciated patients and in *Pandu* (~anemia) etc. It is suitable even in children and old persons [10]. *Shastikashali pinda swedana* was adopted for the case as it is a type of *Abhyanga* and *Mridu swedana* and is suitable for children [11]. It imparts nourishments to the tissues and thus alleviates the *Vataja* disorders. *Niruha basti* is indicated in the treatment of *Udavarta* but it is not suitable in children. Hence, a milder

form of *basti*, *Matra basti* with *Aswagandha taila* was used in initial stage. *Aswagandha taila* is effective in *Vataja* disorder (~neurological, rheumatic, and musculoskeletal diseases) and in emaciation [12]. It also works as a *Rasayana*. In the later stage *Niruha basti* was given as *Erandmooladi yapna basti* as *Yapna basti* is a milder form of *Niruha basti* and can be given in children [13]. This *basti* is indicated in *Chirarsh* (~chronic hemorrhoids). Elsewhere, it is also indicated in *Udara roga* (abdominal diseases which ultimately resulted in ascites), *Mutrakriccha* (~dysuria), *Udavarta* (~mis-paristalsis) and *Gulma* (~abdominal distension) [13]. All these symptoms are similar with *Pakvasayagata vata*. It is indicated in classical Ayurvedic text that *Niruha basti* used in *Udavarta* can *Apanayan* the *Sira* situated at *Guda pradesh* (~nourishment of nerves situated at large intestine and may increases innervations in muscles and thus

increases contractibility of muscles of large intestine) [14]. As *Udavarta* and *Vibandha* (~constipation) are mainly *Vata* predominance disorders, *Brihatavatachintamani rasa* was used as this medicine is indicated in all type of *Vataja vyadhi* (diseases due to *vata dosha*) [15]. *Draksha* (*Vitis vinifera* L.) is having *Anulomak* property. It is the main ingredient of *Drakshaveleha*. *Drakshaveleha* is effective in *Amlapitta* (~hyperacidity), *Kshaya* (~emaciation), *Shotha* (~oedema), *Pandu* (~anemia), *Daha* (burning sensation), *Badhakosthata* (~constipation), *Atisara* (diarrhea), and *Mandagni* (~diminished digestive power) [16]. Hence, this was effective in the case. There is good improvement with treatment in quality of life, bowel distension and constipation in the patient. After the completion of 14 months of treatment patient was able to defecate per day without any discomfort. The treatment is safe for the case as the safety profile of treatment were also within limits. No complication or adverse effects were noticed during the treatment. The case is important one as it throws new light on the possible treatment of HSCR and also help in understanding the pathophysiology and treatment of *Pakvasayagata vata* disease in the context of HSCR. The case report demonstrates clinical improvement in HSCR with *Panchakarma* and Ayurvedic medicinal interventions. There was no need to use any surgical intervention for this case. These *Panchakarma* and Ayurvedic medicinal interventions have the advantage of *Brihman* (~anabolic) effect which is evidenced by weight gain in the boy. So these Ayurvedic intervention is preferable than surgical manipulations as postoperative enterocolitis, incontinence, disrupted anastomosis, fecal fistula, intestinal obstruction and anal stenosis are the main complication in surgical manipulations [17]. As the age of boy advances, there may be some development of new neurons in large colon. At that time patient can be managed with *Mridu virechan* (mild purgation). In the long term patient may be further managed with *Virechan* and *Anulomak* (carminative) foods. For this case long course of treatment and follow up is needed. Experience of this case showed that HSCR may satisfactory be managed with Ayurvedic treatment and controlled trials can be conducted on a good sample size.

8. Conclusion

The case report demonstrates clinical improvement in HSCR with *Panchakarma* and Ayurvedic medicinal interventions.

Patient consent

Written permission for publication of this case study had been obtained from the patient's parent.

Patient's perspective

Parents of the patient were satisfied with the provided treatment.

Sources of funding

None.

Conflict of interest

None.

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