

Case Study

Ayurvedic Pain Management of Sprengel's Deformity: A Case Report

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A B S T R A C T

Introduction: Sprengel's Deformity is a rare congenital disease of unknown etiology characterized by acute pain and disability of the shoulder and neck.

Main Clinical Findings: This is a case report of a 15-years- old female child suffering from pain symptoms and movement restriction of the neck and shoulder.

Diagnosis: Unilateral Sprengel's Deformity Grade II (Ayurvedic diagnosis: Vata-vyadhi).

Interventions: Shrotoshodhan (cleansing of channels), Dosha shaman (balanced state of body humors), Bahya snehana (external oleation), and Swedana (sudation) to nourish Asthi-dhatu (bone tissue), pacify Vata-kapha dosha, and increase the flexibility of Mamsa dhatu (muscle tissue). Yogasanas like Greeva sanchalan (neck movements), Skandha sanchalan (shoulder movements), Shalabhasana (locust pose), and Salamba bhujangasana (Sphinx pose) are advised after reduction of acute pain symptoms.

Outcome: Assessment was done by the shoulder pain and disability index, and improvement was observed in pain and movement restriction.

Conclusion: The case was effectively managed with Ayurveda oral medication, therapy, and Yoga practices.

Keywords: Vata-Vyadhi, Sprengel's Deformity, Greeva Sanchalan, Skandha Sanchalan, Greeva Basti, Kati Basti

Introduction

Sprengel's deformity is a rare congenital abnormality of unknown aetiology, and exhibits dominant transmission. Surgery is advised for complete aesthetic and functional restoration of the shoulder. The scapula appears elevated, protruding in the neck, giving rise to restricted mobility of the shoulder and pain in the cervical spine. Management of Sprengel's deformity depends upon the severity of pain and restriction of movement. Surgery is advised for complete aesthetic and functional restoration of the shoulder.

Patient Information

A 15-years- old female child reported to the outpatient department with her father with chief complaints of shoulder asymmetry, pain in neck, back, left side of the shoulder, and lower back region, difficulty in overhead elevation of the left arm, discomfort in completely stretching the left arm, discomfort, and back pain after sitting continuously for more than thirty minutes for the past two years. She was born full term, appropriate for gestational age, by normal vaginal delivery with spontaneous cry at the hospital. She is the second child of her parents from a non-consanguineous marriage and has two healthy siblings. Her neonatal period went uneventful with normal developmental milestones. The mother noted that the child's good health and muscle bulk caused the asymmetry between her shoulders to go unnoticed until she was two years old. Thereafter, Mother recognised the asymmetry between the two sides and a superiorly positioned left shoulder placed left shoulder. She had complaints of pain in her neck, left shoulder, and lower back, which increased after studying for longer durations in a sitting position for the last 2 years. Pain was insidious in onset, progressive in nature, not associated with numbness, and not radiating to the extremities. There is no family history of minor or major congenital anomalies. In the beginning, the parents went to local pharmacists and dispensaries to get medicines for pain relief; gradually, as the symptoms worsened, they consulted homeopathic, physiotherapy, orthopaedic and neurosurgery specialists and were diagnosed with Sprengel's Deformity. Symptomatic treatment was given for approximately two years; the pain persisted, and the patient was not satisfied with the treatment. The patient came with her parents to seek Ayurvedic treatment for the pain and other symptoms.

Clinical Findings

Upon clinical examination, the patient's vital signs were recorded as follows: temperature 98.8°F, blood pressure 118/72 mm Hg, pulse rate 76 beats per minute, and she was conscious and well orientated to time, person, and place. The left shoulder was elevated above the normal level and lying slightly higher than the right in the standing

position Figure 1. No clear torticollis, no deviation of the head on either side of the neck, and no lumbar deformity like kyphosis, lordosis, or scoliosis were noted. The bilateral size and symmetry of the clavicle appeared normal with normal bilateral acromion processes, and the convexity of the thoracolumbar spine on inspection and palpation was normal. The chest excursion from complete expiration to maximal inspiration was measured at 5 cm, revealing sufficient excursion. The range of motion (ROM) of the left shoulder was found to be restricted, whereas, a normal range of motion was observed in the other shoulder. On the Medical Research Council scale, the right side showed full power (grade 5) of all the muscle groups at the shoulder, whereas the left side showed grade 4. The neurological examination of the lower limbs was unremarkable.

Diagnostic Assessment

Chest-x ray AP view done on 04/11/22 Figure 2, X-ray Cervical Spine AP and lateral view conducted on 05/11/22 reported Figure 3: elevation of scapula, superomedial, and rotated with inferior angle directed laterally with large omo-vertebral bone to cervical spine on left side- F/S/O Sprengel Shoulder (grade II). CT-Spine (16/11/22) had shown fusion of the occipital condyles and anterior arch of the C1 vertebra, reduced C5-C6 intervertebral disc space, and fusion of posterior elements of the C5-C6 vertebrae on the left side. Omo-vertebral bone communicating with C5-C6 posterior elements on the left side with the superomedial aspect of the left scapula along with rotation of the scapula is suggestive of Sprengel Deformity (grade II). Spina bifida is seen from C5 to D1 and D3 vertebrae. Overall findings suggest syndromic congenital disease (D/D Klippel Feil Syndrome). The vitamin D level on May 23 was deficient and was 07.43 ng/mL. X- ray Cervical spine AP and lateral view conducted on May 23 reported bone growth arising from the posterior element of C5 and reaching up to the posterior element of D2. Cervical vertebrae appear normal with reduced disc space at C5 and C6 Figure 4.



Figure 1. Showing Asymmetry Between the Shoulders

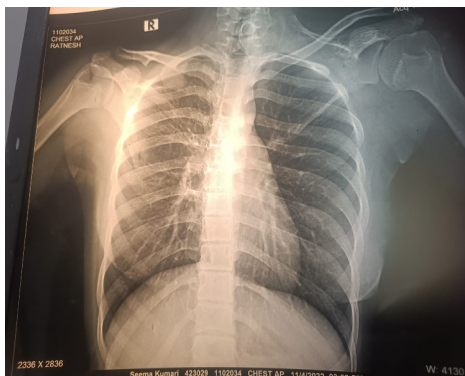


Figure 2.X-Ray Chest AP View (04/11/2022)



Figure 3.X-Ray Spine (05/11/2022)

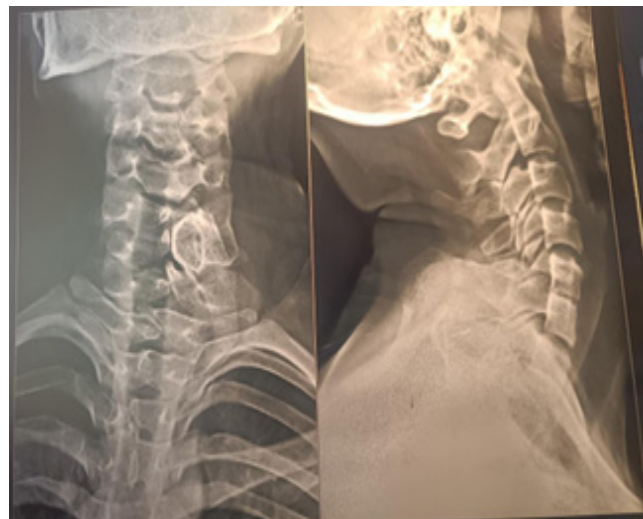


Figure 4.X- Ray Cervical Spine AP and Lateral View (23/05/2023)

Therapeutic Interventions

The patient was thoroughly examined in the outpatient department and advised to take internal medicines for two weeks, along with diet and lifestyle modifications. After two weeks, the patient was admitted to the Inpatient Department (IPD). The timeline of the patient's clinical history, treatment and outcomes are enlisted in Table 1.

Table 1. Timeline of the case before and during Ayurvedic Intervention

Year	Clinical events	Interventions
Uneventful birth and neonatal period (Before 15 years)	The baby was healthy, feeding well, with no observed minor or major congenital anomalies.	—
At an age of 2 years	Asymmetry between two shoulders, with the high left shoulder in comparison to the right.	—
2 years back	Pain in neck, back, and left shoulder.	Oral painkillers by local dispensaries
1 year back	Pain in the neck and shoulder. Cracking sound on movement in the cervical region	Oral painkillers from local dispensaries
29/11/2022	Pain in the neck and shoulders increased. Cracking sound on movement in the cervical region.	Oral homeopathic drugs for 7 days.
		Physical medicine and rehabilitation (PMR): isometric neck strengthening, B/L scapular mobilisation, and TENS at the left periscapular region for seven days
24/12/2022	Continuous pain and cracking sound in the cervical region. Shoulder pain decreased	All the above medicines continue.
		Physiotherapy was continued from 27 to 31 dec 202
17/01/2023	Mild relief, symptoms persist	Oral medicines continued.

15/02/2023	Pain in the neck and shoulders persists. Pain in the middle and lower back region	Oral medicines continued.
12/04/2023	Heaviness in both shoulders, dull pain while raising the arms, and the cracking sound in the neck	Oral continued, and Orthonova Oil was applied locally
23/05/2023	Complaint continued	Oral medicines continued.
06/06/2023	Complaint increased.	Oral drugs:
		Tab Torib MR (4) 1 tab twice daily
		Tab T. Cyra (20) 1 tab once daily
		Tab A to Z 1 tab once daily
		Tab Vernace 1 tab once daily
		Inj. Arachitol (6 Lac) IM Stat
		Cap Vitacef (60 K) 2/week for 3 weeks
		Physiotherapy
06/07/2023	Mild relief in pain, the rest of the illness is regular	Oral medicines cont.
Timeline of the case during Ayurvedic Intervention		
Date of visit	Presentation during different visits	Interventions
OPD 03/08/2023	First visit to the outpatient department -	A combination of: -Aswagandhadi chur-na -1gm Sitoplaladi churna-1gm Godanti bhasma -250 mg Mukta shukti bhasma -250 mg in combination, twice daily with Honey.
	Pain in the neck, back, left side of the shoulder, and lower back region.	Tab Kaishore guggulu (375 mg) 1 tab twice daily with water
	Difficulty in the overhead elevation of the left arm.	Tab Trayadasanga guggulu guggulu (375 mg) 1tab twice daily with water
	Discomfort in completely stretching the left arm.	Tab Rasaraj rasa (125 mg) 1tab twice daily with water
	Difficulty in studying for more than 30 minutes.	
	Discomfort and back pain after sitting for more than 30 minutes	
First follow up to the outpatient department: -		
OPD 10/08/2023	Mild relief from pain	Oral medicines continued
	Other complaints persist	
Date of Admission	Condition of the patient	Intervention
IPD 17/08/2023	Difficulty in raising the hand above the head.	Tab Trayodasanga guggulu guggulu (375 mg) 1 tab twice daily with water
		Tab Lakshadi guggulu guggulu (375 mg) 1tab twice daily with water
	Difficulty in studying for more than 30 minutes.	Cap Abha (600 mg) -1tab twice daily with water
		Cap Arthrum (500 mg) -1tab twice daily with water

	Pain in the shoulder, neck, and back	Madhur virechan churna -2gm once daily with water at bedtime.
		Greeva basti & kati basti for 30 mins with Mahanarayan Tail + Murivenna oil, followed by Nadi swedan with Dash-mool kwath for 15 minutes for 8 days.
Date of discharge 26/08/2023	Easily raise both arms above the shoulders.	Trayodashanga guggulu guggulu (375 mg) 1 tab twice daily with water
		Lakshadi guggulu (375 mg) 1 tab twice daily with water
		Rasnasaptaka kashaya- 20 ml with 1 cup water
	Sitting without difficulty for more than three hours.	Cap Uprise-D3 (60 K) 1 tab per week with milk for 8 weeks
		Cap Abha (600 mg) 1 tab twice daily with water
		Cap Arthrum (500 mg) 1 tab twice daily with water
	Significant relief in shoulder, neck, and back pain.	Madhur virechan churna- 2 gm + Triphala churna- 1 gm once at bedtime with 5 ml of Castor oil and a cup of lukewarm water.
		Local application of Mahanarayan tail
		Yogic practices: Greeva sanchalan, Skandha sanchalana, Shalabhasana, and Sulabha bhujangasana 5 rounds, each for 3 minutes are advised to practice in morning hours.
		Diet and lifestyle modification
07/09/23	Follow up in OPD	Advice given
	Pain symptoms were relieved.	Oral medicines
		Trayodashanga guggulu (375 mg) 1 tab twice daily with water
		Lakshadi guggulu (375 mg) 1 tab twice daily with water
	The range of movement increased.	Rasnasaptaka kashaya- 20 ml with 1 cup water
		Cap Abha (600 mg) 1 cap twice daily with water
		Madhur virechan churna- 2 gm + Triphala churna- 1 gm once at bedtime with 5 ml of Castor oil and a cup of lukewarm water.
	Crepitations or Cracking sound at the level of the neck were reduced.	Local application of Mahanarayan tail pichu (oil tampon)

		Yogic practices: Greeva sanchalan, Skandha sanchalana, Shalabhasana, and Sulabha bhujangasana (7 rounds, each lasting for 4 minutes) are advised to practice in the morning hours.
		Diet and lifestyle modification
03/10/2023	Follow ups in OPD	Same treatment as above. Gradual increase in the duration of Yoga practices.
12/10/2023		
19/10/2023		
21.12.2023	Follow up in OPD	Aswagandha churna -3gm twice daily as processed milk.
		Tab Yogaraj guggulu (375 mg) 1 tab twice daily with water.
		Local application of Mahanarayan tail pichu (oil tampon)
		Yoga practices.

Followup and Outcome

Assessment was done before and after the treatment by using - the Shoulder Pain and Disability Index (SPADI)¹ [Table 2]. The range of movement of the shoulder joint was measured with the help of a goniometer before and after treatment [Table 3]. At the end of the treatment, significant improvement was noted in clinical features

in terms of pain, range of movement, cervical vertebrae, joint crepitations, and cracking sound. After treatment, the SPADI total score improved from 70.75% to 30.375%. Its grade improved considerably from before treatment to after treatment and even more in subsequent follow-ups. Observations had shown encouraging improvement in the pain and other symptoms [Table 4].

Table 2. Shoulder Pain and Disability Index (SPADI)

Pain Scale	Before Treatment	After Treatment	3 rd Follow-Up
At its worst?	8	6	4
When lying on the involved side?	10	8	5
Reaching for something on a high shelf?	8	5	3
Touching the back of your neck?	8	5	2
Pushing with the involved arm?	8	4	2
Total points	42	28	16
Total pain score (%)	84	56	32
0 = no pain and 10 = the worst pain (0-to-10-point imaginary pain scale) (Total pain score in % = Total Points/ 50 x 100)			
Disability Scale	Before Treatment	After Treatment	3 rd Follow-Up
Washing your hair?	5	3	3
Washing your back?	8	5	3
Putting on an undershirt or jumper?	5	3	3

Putting on a shirt that buttons down the front?	5	3	3
Putting on your pants?	3	3	3
Placing an object on a high shelf?	10	5	3
Carrying a heavy object of 4.5 kilograms.	5	3	2
Removing something from your back pocket?	5	3	3
Total Points	46	28	23
Total Score (%)	57.5	35	28.75
0 = no difficulty and 10 = so difficult it requires help. (0-to-10-point imaginary disability scale) (Total disability score % = Total Points / 80 x 100)			
Total SPADI score= Average of Total pain score & Total disability score BT- 70.75 %, AT- 45.5 %, FU- 30.375 %			

Table 3. Range of movement of affected Shoulder joint

Range of Movement (Normal range)	Before Treatment	After Treatment
Shoulder Flexion (180°)	120°	160°
Shoulder Extension (45°-60°)	50°	50°
Shoulder Abduction (150°)	110°	140°
Shoulder Adduction (30°-50°)	30°	40°
Medial Rotation (70°- 90°)	70°	70°
Lateral Rotation (90°)	70°	80°

Table 4. Improvement in the pain and other clinical features.

Features	Before treatment	After treatment
Symmetry between shoulders	Asymmetrical (Left shoulder higher than the right)	Asymmetrical (unchanged) (Left shoulder higher than the right)
Pain Symptoms	Pain in the neck, shoulders, and back.	Pain relieved
Cracking sound	Present on an upward-downward, left-right rotation of the head.	Absent
Vertigo	Not present	Not present
Hour of sitting without discomfort/ pain	30 minutes	3-4 hours

Discussion

There is a concept of Janmabala pravrita vyadhi (congenital disorders) responsible for the development of Pangu (disabled body part).² Also, the injuries occurring during birth are responsible for the development of Vikar (abnormal size, shape, or placement) body parts.³ Vitiating of Vata in foetus Kubja (hunch backed), Pangu (crippled), Muka (dumb) etc.⁴ Even though it is impossible to cure the disease, symptoms can be managed to improve the quality of life. The symptoms of the diseases, like pain, stiffness, restricted movement of the shoulder, crackling sound, discomfort in sitting positions, etc., are mainly due to the involvement of Vata and Kapha. The function of Vata is to provide Gati (movement); the vitiated Vata restricts the movement. Here, the treatment is aimed at nourishing Asthi-dhatu (bone), pacify Vata-kapha dosha, increase the flexibility of Mamsa dhatu (muscle), and maintaining the function of shoulder and cervical vertebrae to do daily chores without any discomfort. As the pain symptoms dominate, the treatment was started with Rasaraj Rasa⁵, Trayadasanga guggulu⁶, Kaishore guggulu⁷ and Aswagandhadi churna⁸, Sitopalaladi churna⁹, Godanti Bhasma¹⁰ (Calcium sulphate), and Mukta shukti Bhasma¹¹ in combination to prevent further Dhatukshaya. Rasaraj rasa contains gold, silver, and iron Bhasma, which balance the Vata and give strength to nerves and muscles. Trayodashanga guggulu is useful in various Vata disorders (neurological, rheumatic, and musculoskeletal diseases). Kaishore guggulu acts on Mamsa dhatu. Apart from the muscle strengthening and rejuvenating activity of Aswagandha (*Withania somnifera*), experimental studies support the skeletal muscle relaxant activity of the aqueous extract of *Withania somnifera* roots in albino mice.¹² Godanti bhasma has Shoolahara (analgesic) and Balya (tonic) properties, it is a source of natural calcium.

After two weeks of oral medication, the patient was admitted to perform Greeva basti and Kati basti with Mahanarayan Oil¹³ and Murivenna Oil in an equal ratio.¹⁴ The procedure Greeva basti and Kati basti (external oleation) are significant in the Shaman of Vata Dosha, because they bring Mriduta (softness) to the hardened body tissues and relieve restricted neck movement. Following Basti, the affected part was given Nadi sweda which further alleviates the Vata dosha and increases the flexibility of the muscles. Lakshadi guggulu has antiarthritic and chondroprotective activity. As the bowel habit of the patient was not regular and to give Nitya virechana (regular purgation) to relieve Shrotorodha, a combination of Madhur Virechan churna and Triphala churna was advised once at bedtime with castor oil and a cup of lukewarm water. At the time of discharge from the hospital, when the acute symptoms subsided, along with oral medicines, Yogic practices like Greeva

sanchalan, Skandha sanchalana, Shalabhasana and Salamba bhujangasana were advised to maintain the flexibility of muscles and movement of joints. Salamba bhujangasana (Sphinx Pose) is effective for chronic low back pain. During Bhujangasana, the erector spinae and transverso-spinalis muscles show powerful contraction and strengthening; the serratus posterior superior muscles are involved in chest expansion and internal arm rotation; thoracic back flexion, and stability are performed by the latissimus dorsi. In Shalabhasana, the middle trapezius performs lateral rotation in the prone arm raise, the latissimus and the rhomboids are involved in arm and upper back lengthening. Studies show a reduction in work related chronic neck pain, relief of muscular tension around the neck-shoulder region by improving cervical range of motion, increased neck flexibility, and muscles strengthening effects of Greeva sanchalan. Skandha sanchalana had been observed as an adjunct therapy in the management of mild to moderate cervical spondylosis. Therefore, it's practice enhances both the toning of muscles and the release of muscle tension. The induced relaxation reduces muscle tension and modifies neurobiological pain perception.

Conclusion

No absolute medical management is available for Sprengel's deformity. Ayurvedic intervention is a ray of hope for improving the functional status and quality of life of the patient in cases of congenital deformities where there is no confirmatory treatment available other than surgical correction. Clinically significant improvement is observed in the patient with Ayurvedic management (including outpatient and inpatient management). Regular and prolonged followups of the case are needed for further evaluation.

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