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Management of Hirayama Disease through *Panchakarma*- A Case Report

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ABSTRACT:

Hirayama disease (HD) also called as Monomelic amyotrophy (MMA) is characterized by juvenile onset of unilateral muscular atrophy of a distal upper extremity. The pathogenic mechanism of Hirayama disease is cervical cord compression by the posterior dura with forward displacement in the neck flexed position. Here is a case report of a 26 year old male with HD who had been treated at outpatient and inpatient level of VPSV Ayurveda hospital. The case was considered as a *Vatavyadhi* and the treatment protocol applied accordingly with incorporation of treatment of *Apabahuka* particularly. The treatment consisted of a variety of treatments including *Udvartana*, *Snehapana*, *Swedana*, *Virechana*, *Vasthi*, *Nasya and Rasayana*. Even though the condition of the patient was irreversible, the patient could hinder the course of disease during follow up and was able to regain the functional status after restarting the treatment later. The case highlights the improvement in quality of life of Hirayama patient along with improvement in UEFI score after Ayurvedic management.

Keywords: Hirayama disease, Monomelic atrophy, *Vatavyadhi*, *Apabahuka*, *Panchakarma*

INTRODUCTION

Hirayama Disease (HD) is a rare neurological entity characterized by self-limited asymmetrical slowly progressive atrophic weakness of forearms and hands, either unilateral or bilateral and without sensory loss.[1,2] The disease Hirayama was first described by Keizo Hirayama as a juvenile muscular atrophy of unilateral upper extremity. For many decades, the condition was thought to be unilateral and sporadic one with idiopathic causes. Though the pathogenesis is unknown, probable causes suggest that an imbalanced growth between the vertebral column and spinal canal contents. Current neuroradiologic techniques are able to show forward displacement of the posterior wall of the lower cervical dural canal in neck flexion, which is presumed to be a primary pathogenetic mechanism of Hirayama disease. The Motor Neuron Disease is a very close differential diagnosis of HD but the peculiar course of initial progression and spontaneous arrest in the later period is seen in the latter. It can cause significant disability in some subsets of individuals and early interventions can minimize the affliction.[2] Here we report a case of HD who was treated with Ayurvedic interventions and shown encouraging results.

Patient Information

A 26 year old male patient reported to outpatient department with weakness of bilateral upper limbs (Lt>Rt)

and unable to raise hands for the last 8 years. The patient was apparently healthy till 12th standard. He was interested in exercising since childhood and practiced all kinds of sport activities such as weight lifting, cricket etc since 6th standard. He had started practicing martial arts since 10th standard, and used to perform all Yogaasana, including seershasana as a part of exercises. While he was in 12th standard, he felt tiredness during exercises. Gradually he noticed slight wasting of left upper limb muscles along with the prominence of shoulder joint. He also developed weakness of right upper limb and felt difficulty in push up, lifting weight and lose in holding capacity. He noticed occasional tremor of fingers. Routine laboratory investigations were within normal limits. While undergoing evaluation MRI showed cervical disc prolapse at C4-C5 and C5-C6 (Fig.1). Nerve conduction studies showed asymmetrically reduced CMAP amplitude from left musculocutaneous, axillary and suprascapular nerves. Electromyography showed active and chronic denervation and reinnervation from cervical and lumbosacral myotomes (Fig.2). The patient received the diagnosis of Hirayama Disease or Monomelic amyotrophy (MMA) based on the clinical, electrophysiological and radiological characteristics. He took allopathic medications for around one year and got no significant improvement. Then he was advised to undergo surgical intervention and since he was reluctant to undergo surgery he came for ayurvedic treatment, first from outpatient department and then he got admitted in the inpatient department for better management. He had no significant past medical and surgical histories. No history of exposure to toxins/allergies/addictions reported. Family history was not significant.

Neurological examination revealed a fully conscious, alert with normal higher mental functions and cranial nerves. Motor examination showed wasting and reduced power (G4/5) in left upper limb muscles, thenar and hypothenar eminences with sparing of brachioradialis. Tone was normal. Deep tendon reflexes also found to be normal all over.

The patient was assessed with Upper Extremity Functional Index (UEFI), which is used to measure the functional impairment in cases of upper limb musculoskeletal dysfunction. It is a patient reported outcome measure which consists of 20 questions on a 5- point rating scale assessing level of difficulty in performing activities of daily living using the upper extremities including household and work activities, hobbies, lifting a bag of groceries, washing your scalp, pushing up on your hands, driving etc.[3] Before the treatment, it was 73 in this case (Table 1).

Management

Combining the principles of management of *Apabahuka, Bahusosha, Avruthavatha and Sahaja roga*, the treatments were formulated consisting of both external and internal medications (Table No.2). The patient completed the first schedule of *Sodhana* procedures (from *udwartana to vasti*) by 40 days. In the treatment, three days of *Rasayana* was done in the hospital, later he was discharged with an advise to continue the medicine from home itself. Even though no significant change was noticed there were no deterioration in the condition and the patient got a subjective feeling of wellbeing after the treatment. UEFI remained as 73. After one year the patient was again admitted in the hospital and underwent treatments (Table No.3). He was on regular telephonic contact and necessary advises were given. After a few months we lost the follow up of the patient and later came to know that he went for another medical advice and was under their work up. The patient returned to us after 7 years. It was seen that the disease has progressed considerably, he developed more restrictions in the daily activities and the UEFI score was 45. He was treated at outpatient level with internal medications and had regular follow up for 5 months. The medicines consisted of *Anulomana* followed by *Rasayana. Pratimarsha nasya* was also continued (Table No.4). After 3 months the UEFI score showed an increase up to 49 which further increased up to 55 and 59 (Table No.1).

DISCUSSION

HD is a benign form of chronic focal amyotrophy of cervical myotomes especially C7, C8 and T1 segments. [4] Even though it was considered as a rare disease when it was identified, HD cases are now being reported more frequently. HD is reported to have an insidious onset, slow progressive and self-limiting course with subsequent clinical stabilisation in 3-5 years.[5] No definite treatment has been reported to cure the condition and the only hope is its self- limiting nature. But many patients suffer a lot with the functional compromise created by the disease and hence any system giving an improvement in the condition is worthwhile. The present case showed significant improvement in the course. Different approaches are seen in treating HD with Ayurveda. In a case report by *Nandigoudar* et al the condition was correlated to *Visvachi* whereas Pooja Sarma et al correlated to *Asthimajjagata vata*.[6,7] But in this case *Avarana* has been considered leading to wasting of muscles. However the pathological

sequence may ultimately end in *apabahuka or bahususha* hence their treatment also play role in the management of HD. Obstructions in the passages can result in the desiccation of *rasadi dhatu*.[8] The diseases with atrophy of muscles are seen to be critical like that of Motor neurone disease hence a long term treatment plan was made in the present case with a prior importance to the pathology of *Avarana*. In the first course *udwartana and takrapana* were advised as initial therapies which were intended for *rookshana* purpose and to reduce *kaphavarana*. The qualities of *Takra* such as *laghu guna, amla rasa and deepana* property ensures its actions.[9]

After *rookshana*, *sodhananga snehapana* was done with *Mahamasha taila* which is specifically indicated in conditions affecting upper limbs like *apabahuka, viswachi* etc.[10] As it was assumed to be a disease due to *kaphavata, taila* was selected for *snehapana. Swedana* followed by *virechana and samsarjanakrama* administered as per their conventional method. *Prasarani taila,* which is indicated for all *vatavyadhi,* was administered as *anuvasana and madhutailikavasthi* was administered as *niruha in yogavasthi*. Owing to the incurability and *kaphavarana* nature *silajatu* was preferred as *rasayana.*

Internal medications administered before *rasayana* were *Gandharvahastadi Kashaya, Chiruvilwadi Kashaya, Vaiswanara chūrna, Hinguvachadi chūrna, and pippalyasava*.[11] The common factor in the medicine selection was *deepana* property which aimed at *Rasadhathwagni vardhana. Rasadhatu vaigunya* has a main role in this disease pathogenesis. The common trend of alleviation of *kapha and vata* was also continued in the follow up medicines and thus formulations like *Intukanta, shatpala* etc were advised.

The treatment protocol planned for the second course consisted of treatment of '*Vatavyadhi'* with incorporation of *Apabahuka chikitsa*. *Snehana, swedana and sodhanam* are the general treatment principles of *Vata.* Hence *snehapanam, abhyanga-ushma sweda* and subsequent *Virechana* and then *Vasti* was planned. In short the schedule consisted of *brimhana* therapies mainly with needful *sodhana* therapies in between them. *Snehapana* was done with *Rasaghrutham* which is specifically mentioned for prevention of quick spread of Vata all over the body.[12] The ingredients of *Rasaghruta* such as *Vidaryadi gana, ksheera, Dadhi and Mamsarasa* strengthens the nerves and also have *vatahara* properties.[12] *Lakshadi taila* was used for *Abhyanga* after *snehapana* which is ideal for this condition such as '*Balyam', 'Kshyayam' and 'Vatanut'*.[13] *Ushmasweda* also carried out after *Abhyanga* with *Lakshadi taila,* which was aimed for elimination of bodily wastes from *sakha to koshta*. For *virechana,* the safest at the same time having property of curing *kapha* was meant and thus *vidangatanduladi churna* was selected.[14] Following the principle of repeated administration of *snehana and sweda for vata, pizhinjuthadaval* done which gives *snehana* and *swedana* simultaneously. The medicines used were *Panchasneha and Lakshadi kuzhambu. Panchasneha*, is indicated in *Bahususha, Apabahuka,Bahustambha and Vataroga.* In addition to that, it also causes *Bahuvruddhi.*[15] Towards the end of *pizhinjuthadaval, yogavasthi* was planned to fulfil purification of the body. *Matra vasthi* was administered with *Nimbamruthadi eranda tailam. Doshotklesa or sodhana* property of *nimbamrutadi eranda taila* is considered here for the selection as *matravasti dravya*. After two days of administration of *matravasti, madhutailikavasthi* was administered for the next two days and *matravasti* also administered in the afternoon on the same days, to control *vata.* After two instillations of *madutailika vasthi, doshahara vasthi* was planned for one day along with which *matravasthi* also administered on the very same day afternoon. The dosha which became ready to expel by *matravasthi* was eliminated through *Doshaharavasti* and after that, *Mustadi rajayapana vasti* was administered for the next two days which is *mamsa-agni vardhana and dhatuposhana*. Several studies shown the effect of *Mustadi rajayapanavasthi* in improving physical status.[16] As it is *brumhana* in properties, the *matravasthi* administered on those days also planned as *brumhana and Mahamashataila* was selected for *matravasthi*..The most commonly encountered *brimhana* therapy, *Shashtika shali pinda Sweda* (SPS) was administered to make further improvement in muscle bulk. Its ingredients like milk and *Shashtika Shali* nourishes and gives strength to muscle tissues. *Bral* fish(Snake head fish) meat used along with *shashtika pinda* for the better outcome of the procedure. As per the recent studies, bral fish meat helping to fix tissue damage and it is a natural source of plenty of albumin, which has an important role in improving cell regeneration process. Also, the high protein content would help a lot in the process of formation of muscles. *Lakshadi* and *balaswagandhadi taila* were used for abhyanga purpose during SPS. *Balaswagandhadi taila* cures different kinds of *vatarogas* and explained as one among the best to produce *pusti.*[17]

Nasyakarma using *Rasataila* was the final procedure administered during the period of IP treatment, which was also aimed to improve the strength of shoulder muscles and

administered for 5 days with multiple instillations in the same sitting (ie, 2 ml, 2+2ml and 2+3ml. *Rasataila* is a modified form of *Karpasasthyadi taila* added with head meat of goat. It is *brumhana, mamsavardhana* and indicated in *urdhwajatrugata vikaras and samsargaja vatavyadhis.*

Indukantham kashayam and *ghritham* were administered internally as it is specifically indicated in *Vatamaya* and *kshaya.* Also it is having the properties of *balavardhana, agnideepana and srotosodhana. Balarishtam and partharishtam* were administered as a combination which focussed on correction of functions of *vyanavata*. 'Rasa dhatu ayana' is the function of *vyanavayu* which situated in *hrudaya* and correction of *vaigunya* is needed for the *samprapti vighatana.*

After the two courses of IP level management, condition of the patient was much better and quality of life also improved. But after the first outbreak of Covid-19 pandemic, patient couldn't continue the medications and then he restarted the OP level medications in 2021 for improving his general conditions. Patient was comfortable with the medications. *Pratimarsa nasya and rasayana* were considered in OP level management for a long-term benefit. *Shadbindutaila* quoted in BRV *Sirorogadhikara* was given for *pratimarsa nasya* which is specifically indicated in *bahubalakshaya.* It provides *adhika bahubala* as per the *phalasruthi* which is needed for the condition of the patient. Prior to the administration of *Narasimhachurna* as a *rasayana, vidangatanduladi churna* was advised as *sadyovirechana* to ensure the cleaning of channels.

On the second visit (after 3 months), patient was advised to do *virechana* with modified *Katukamalakadi kashaya*.[18] Half of the ingredients from the original yoga were used for *virechana* purpose i.e., *katukarohini, amalaki and guduchi.* After *virechana* he was advised to take *sarvamayanthaka ghritha* in capsule form at bedtime. *Sarvamayantaka ghrita* also cures all kinds of diseases and has a specific indication in *'karasthambha'* which shows its action on upper limbs.[19] After the initial administration of *shadbindu taila* for 3 months as a *sodhana nasya,* the procedure was changed to *brumhana* line and *rasataila* was administered which is better in *urdhwajatrugata vikaras.*

Even though improvements were noticed in quality of life, body weight and UEFI Scale, patient still has some difficulties related to the movements of upper limbs as the disease is chronic and has no complete cure for the condition. As the condition is *yapya* in nature, the entire treatment protocol is aimed to manage the condition without further progression of the disease and to offer the better quality of life to the patient.

CONCLUSION

Patient was being treated with a *Vatavyadhi* based protocol both in IP and OP levels with incorporation of *Apabahuka chikitsa* in conditions where necessary. A few important formulations and treatments were also used accordingly. A significant improvement is noticed in this case with the above mentioned treatments. Though there was a gap between the IP-OP treatments due to the pandemic issues etc, the role of this specific treatment protocol in disease history is clear. The ongoing OP level treatment also seen as beneficial to the patient as recognizable improvement was seen in the UEFI scale. It is evident that Ayurveda line of management is effective in managing chronic neurological conditions like Hirayama and the same will be further proven in the future with clinical trials.

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Table No.1 Upper Extremity Functional Score

No	Item	Score BT	Score After 7 years	Score after 7 years-follow up 1	Score after 7 years-follow up 2	Score after 7 years-follow up 3
1.	Any of your usual work, housework, or school activities	4	3	4	4	4
2.	Your usual hobbies, recreational or sporting activities	3	2	3	3	3
3.	Lifting a bag of groceries to waist level	3	1	2	3	3
4.	Lifting a bag of groceries above your head	2	1	2	2	2
5.	Grooming your hair	4	2	2	3	3
6.	Pushing up on your hands (eg, from bathtub or chair)	3	1	1	1	1
7.	Preparing food (eg, peeling, cutting)	4	2	2	2	3
8.	Driving	4	3	3	3	3
9.	Vacuuming, sweeping or raking	4	2	2	3	3
10.	Dressing	4	3	3	3	3
11.	Opening doors	4	3	3	3	3
12.	Cleaning	4	3	3	3	3
13.	Tying or lacing shoes	3	2	2	2	3
14.	Sleeping	4	4	4	4	4
15.	Doing up buttons	3	3	3	3	3
16.	Using tools or appliances	4	2	2	3	3
17.	Laundering clothes (eg, washing, ironing, folding)	4	2	2	3	3
18.	Opening a jar	4	2	2	2	2
19.	Throwing a ball	4	2	2	2	3
20.	Carrying a small suitcase with your affected limb	4	2	2	2	3
	Total	**73**	**45**	**49**	**55**	**59**

Table No.2 Inpatient treatment details during first course

Internal medications	**External therapies**
Gandharvahastadi ks 90 ml bd (6am&6pm)	*Udvarthana with Yavakolakulatha churna*-7 days
Vaiswanara churnam 5g with hot water bd	*Takrapana*-5 days
Chiruvilwadi ks 90 ml bd (6am&6pm)	*Snehapana* with *Mahamasha taila* -7 days
Hinguvachadi churnam 1 tsp with hot water bd	Baspasweda-3 days
Pippalyasava 25 ml bd a/f	*Virechana* with *Nimbamruthadi erandataila*-1 day
	Anuvasana vasthi with Prasaranyadi taila
	Madhutailika vasti – 3 days
Advice on discharge	*Silajatu rasayana*-30 days
Intukantham ks + Shadpala ghritam – 6am & 6 pm *Balaswagandhadi + Panchasneham* -for external	

Table No.3 Treatment details during second course

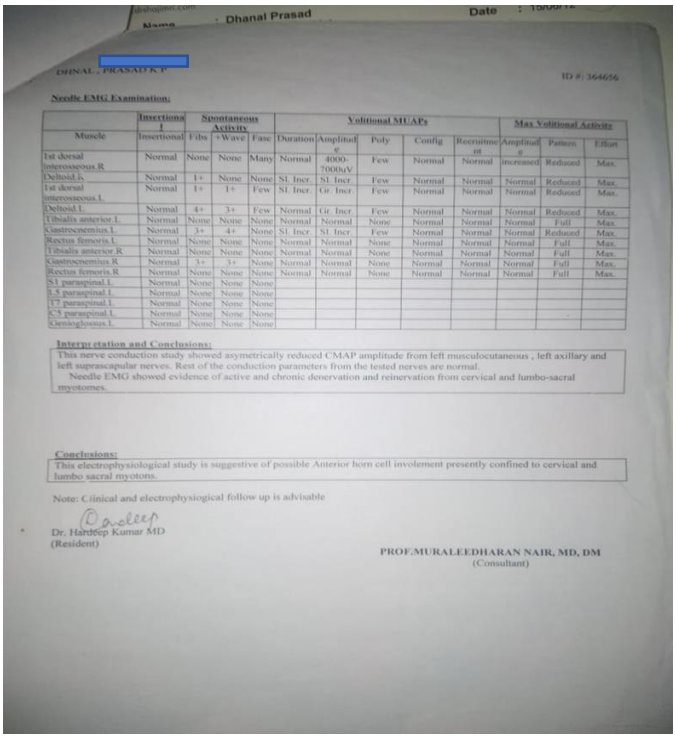
Internal medications	**External therapies**
Intukantham ks 75 ml bd (6am & 6pm) – 2wks	1. *Takrapana* – 4 days
Shaddharana churnam 5g bd with hot water	2. *Snehapana with Rasaghrita* (30, 60, 80, 100, 120, 150, 180, 210 ml)
	3. *Abhyanga-ushmasweda – 3days*

	(Lakshadi taila)
	4. Virechana with Patolamooladi ks 75 ml + Vidangatanduladi churnam 20 g (8am)
Intukantham ks with 1 tsp intukantha ghritam bd (6am & 6pm)	5. Pizhinjutadaval with Panchasneha + Lakshadi kuzhambu – 7 days
Balarishtam + Partharishtam 25 ml twice daily	6. Matravasthi with Nimbamrutadi eranda tailam – 4days
	7. Madhutailika vasthi – 2 days
	8. Doshahara vasthi – 1 day
	9. Matravasti with Mahamashataila – 8 days
	10. Shashtika pinda sweda with Bral fish meat– 7 days Lakshadi + Balaswagandhadi taila for abhyanga
	11. Nasya with Rasataila – 5 days (2ml, 2+2 ml, 2+3ml)
Advice on discharge	
Prasaranyadi kashayam 75 ml twice daily	
Mahakalyanaka ghritam 10 ml at bedtime	
Mahamasha taila + Balaswagandhadi taila for external application	
Asanavilwadi taila – for head	

Table No. 4 Treatment details of third course

Follow up	**Medications**
After 7 years	• *Inthukantham ks 75 ml bd* • *Shadbindu tailam 2drops/ 3 times a day* • *Vidangatanduladi churnam 20 g wih hot water*
First	• *Katukurohini + amalaki + guduchi ks (One day)* • *Narasimha churnam 10 g with milk at night*
Second	• *Cap. Sarvamayanthakam 2 HS* • *Rasatailam 3 drops/nostril 2 times*

Fig.1 MRI of Cervical spine



Name : Dhanal Prasad Date : 15/06/12

DHNAL . PRASAD K P ID #: 364656

Needle EMG Examination:

Muscle	Insertional	Fibs	+Wave	Fasc	Duration	Amplitude	Poly	Config	Recruitment	Amplitude	Pattern	Effort
	Insertional Activity		Spontaneous Activity		Volitional MUAPs					Max Volitional Activity		
1st dorsal interosseous R	Normal	None	None	Many	Normal	4000-7000uV	Few	Normal	Normal	increased	Reduced	Max.
Deltoid L	Normal	1+	None	None	Sl. Incr.	Sl. Incr.	Few	Normal	Normal	Normal	Reduced	Max.
1st dorsal interosseous L	Normal	1+	1+	Few	Sl. Incr.	Gr. Incr	Few	Normal	Normal	Normal	Reduced	Max.
Deltoid L	Normal	4+	3+	Few	Normal	Gr. Incr.	Few	Normal	Normal	Normal	Reduced	Max.
Tibialis anterior L	Normal	None	None	None	Normal	Normal	None	Normal	Normal	Normal	Full	Max.
Gastrocnemius L	Normal	3+	4+	None	Sl. Incr.	Sl. Incr.	Few	Normal	Normal	Normal	Reduced	Max.
Rectus femoris L	Normal	None	None	None	Normal	Normal	None	Normal	Normal	Normal	Full	Max.
Tibialis anterior R	Normal	None	None	None	Normal	Normal	None	Normal	Normal	Normal	Full	Max.
Gastrocnemius R	Normal	3+	3+	None	Normal	Normal	None	Normal	Normal	Normal	Full	Max.
Rectus femoris R	Normal	None	None	None	Normal	Normal	None	Normal	Normal	Normal	Full	Max.
S1 paraspinal L	Normal	None	None	None								
L5 paraspinal L	Normal	None	None	None								
T2 paraspinal L	Normal	None	None	None								
C5 paraspinal L	Normal	None	None	None								
Genioglossus L	Normal	None	None	None								

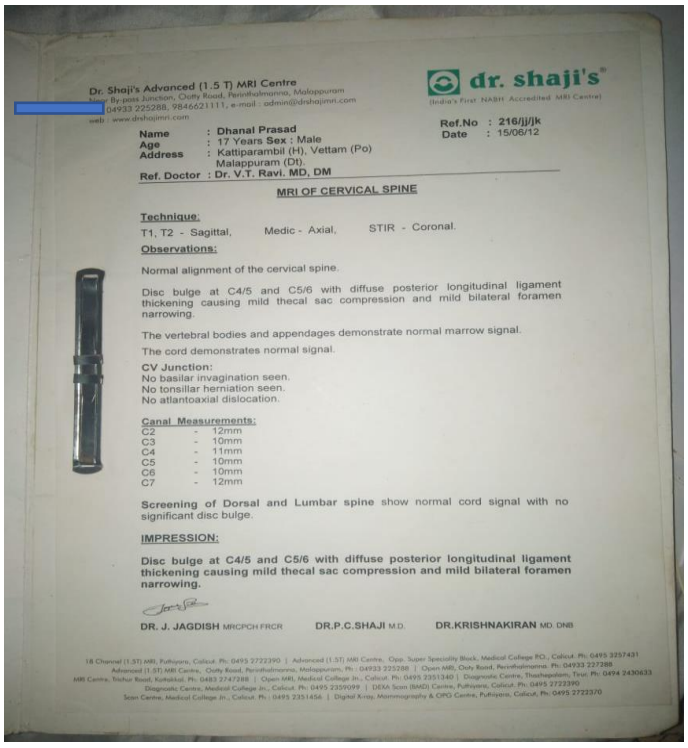
Interpretation and Conclusions:
This nerve conduction study showed asymetrically reduced CMAP amplitude from left musculocutaneous , left axillary and left suprascapular nerves. Rest of the conduction parameters from the tested nerves are normal.
Needle EMG showed evidence of active and chronic denervation and reinervation from cervical and lumbo-sacral myotomes.

Conclusions:
This electrophysiological study is suggestive of possible Anterior horn cell involement presently confined to cervical and lumbo sacral myotons.

Note: Clinical and electrophysiogical follow up is advisable

Dr. Hardeep Kumar MD
(Resident)

PROF.MURALEEDHARAN NAIR, MD, DM
(Consultant)



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dr. shaji's
(India's First NABH Accredited MRI Centre)

Ref.No : 216/jj/jk
Date : 15/06/12

Name : Dhanal Prasad
Age : 17 Years Sex : Male
Address : Kattiparambil (H), Vettam (Po) Malappuram (Dt).
Ref. Doctor : Dr. V.T. Ravi. MD, DM

MRI OF CERVICAL SPINE

Technique:
T1, T2 - Sagittal, Medic - Axial, STIR - Coronal.

Observations:
Normal alignment of the cervical spine.
Disc bulge at C4/5 and C5/6 with diffuse posterior longitudinal ligament thickening causing mild thecal sac compression and mild bilateral foramen narrowing.
The vertebral bodies and appendages demonstrate normal marrow signal.
The cord demonstrates normal signal.
CV Junction:
No basilar invagination seen.
No tonsillar herniation seen.
No atlantoaxial dislocation.

Canal Measurements:
C2 - 12mm
C3 - 10mm
C4 - 11mm
C5 - 10mm
C6 - 10mm
C7 - 12mm

Screening of Dorsal and Lumbar spine show normal cord signal with no significant disc bulge.

IMPRESSION:
Disc bulge at C4/5 and C5/6 with diffuse posterior longitudinal ligament thickening causing mild thecal sac compression and mild bilateral foramen narrowing.

DR. J. JAGDISH MRCPCH FRCR DR.P.C.SHAJI M.D. DR.KRISHNAKIRAN MD. DNB