



## IJPMPR 2013 December; Volume 24 (Number 4)

### Contents

#### Editorial

International Day of Persons with Disabilities. RN Haldar.

#### Original Article

I) Urodynamic study of Bladder Behaviour in Traumatic Spinal Cord Injury Patients in Response to Rehabilitation. Singh Ningthoujam Jungindro, Keshkar Sanjay, Singh Naorem Ajit, Kumar Ratnesh.

II) Efficacy of Diacerein in the Treatment of Osteo-arthritis of Knee. Loitongbam L Sushil Singh, Handa Gita, Singh U, Yadav SL.

III) Intra-articular Steroid in the Management of Adhesive Capsulitis of Shoulder: A Comparison of the Anterior and Posterior Approaches. Sharma G Sonachand, Nandabir Singh Y, Bimol Singh N, Touthang Alex T, Tamphaleima KH

#### Case Report

I) Electrodiagnosis of Quadrilateral Space Syndrome. Dholakia Madhuri, Sinha Anupam.

II) A Multidisciplinary Rehabilitation Approach for Writer's Cramp. Tripathi D R, Kumar Amod, Talele Mahesh, Singh Suryabhan.

III) Plexiform Neurofibromatosis with Abnormal Pachydermatocele: A Rare Case Report. Agarwal AK, Gupta Anil Kumar, Kumar Dileep, Kumar Vijay, Yadav Ganesh.

#### Pictorial CME

Erythema Multiforme in a Case of Polyarthrititis. Pramanik R, Das P.

#### PG Forum

A) Rehab Challenges

B) Book News

C) Article News

D) Rehab Quiz

#### Medical Philately

#### Obituary

Dr Krishna Kumar Singh.

#### Index

Authors and Reviewers

#### Editorial Board

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## Authors' Index of Volume 24, IJPMR

Abdulgafoor S	19	Kumar Amod	104	Sahoo P K	16
Agarwal A K	110	Kumar Deepak	51	Sathyamoorthy R	3
Ameed Equebal	70	Kumar Dileep	110	Senthilvelkumar T	76
Anil Kumar G	44	Kumar Ratnesh	87	Sharma G Sonachand	99
Balamurugesan Kandan	56	Kumar Vijay	110	Singh Naorem Ajit	87
Ballav Ambar	70	Laisram Nonica	63	Singh Ningthoujam Jungindro	87
Bimol Singh N	99	Loitongbam L Sushil Singh	92	Singh Suryabhan	104
Borah Diganta	63	Mansoor S N	29, 80	Singh U	9, 92
Chandy B R	23	Meena N	3	Sinha Anupam	104
Chanu A R	40	Menon Nitin A	73	Soumya V	19
Das P	114	Mohandas Kurup V K	3	Sreekumar V	73
Das S P	16	Mohanty Ram Narayan	16	Sumalatha K B	9
Dholakia Madhuri	104	Mohes A S	27	Talele Mahesh	104
Ghosal Vasundhara	70	Muzaffar Tufail	63	Tamphaleima KH	99
Gogia Virinder Singh	51	Nandabir Singh Y	99	Th Khelendro Singh	27
Gupta Anil Kumar	110	Neyaz Osama	70	Tharion G	23
Handa Gita	9, 92	Ngampa Sangme	27	Touthang Alex T	99
Harshanand P	44	Nilachandra Singh L	27	Tripathi D R	106
Hmingthanmawii	40	Pertin M	40	Viswanathan Stalin	56
Jayasree R	44	Pramanik R	114	Vivek P	44
Jhalani Rakesh	70	Raji Thomas	76	Wadhwa R K	63
Joy Singh A K	27	Ramesh S	3	Yadav Ganesh	110
Keshkar Sanjay	70, 87	Rathore F A	29	Yadav S L	92
Kothari S Y	63, 73	Romi Singh N	40	Zonunsanga C	40
Krishnaprasad I N	19	Sahoo J	16, 48		

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# Efficacy of Diacerein in the Treatment of Osteo-arthritis of Knee

Loitongbam L Sushil Singh<sup>1</sup>, Handa Gita<sup>2</sup>, Singh U<sup>3</sup>, Yadav S L<sup>4</sup>

### Abstract

Osteo-arthritis, the most common joint disease of human being, presents with pain and stiffness of the affected joints, feeling of instabilities, deformities and severe loss of function of the involved joint affecting activities of daily livings as well as economic burden.

The present study evaluates the efficacy and adverse effects of diacerein, an anti-inflammatory drug now being considered as a disease modifying drug in osteo-arthritis on 38 patients. After 6 weeks of intervention, diacerein showed statistically significant superiority versus aceclofenac as assessed with WOMAC A ( $P < 0.0001$ ), WOMAC B ( $P < 0.0001$ ), WOMAC C ( $P < 0.0001$ ) and secondary efficacy variable like NSAIDs (aceclofenac) intake from the baseline and at the efficacy time point at 6 weeks and 12 weeks ( $P < 0.0001$ ), demonstrating the better efficacy of diacerein over aceclofenac. This superiority was already evident from 6th week for all the parameters. It was more significant after completion of the study.

**Key words:** Osteo-arthritis, diacerein, WOMAC, aceclofenac, Kellgren- Lawrence grading.

### Introduction:

Studies by various investigators over the last few decades have demonstrated the inflammatory component in the pathogenesis of osteo-arthritis. Diacerein, a drug which inhibits the pathways and metabolism of inflammatory intermediates interleukin-1 and tumour necrosis factor in various studies<sup>1,2</sup> have shown diacerein to be as efficacious or even better than the commonly used analgesics and NSAIDs in the treatment of both knees without the unwanted side-effects of these medications, with a carryover effect of

its benefits, which according to some studies extend up to 3 months. The present study has shown positive results of the efficacy and safety of the mentioned drugs.

### Aetiology and pathophysiology of osteo-arthritis:

Osteo-arthritis may be idiopathic, where causes are not identified, or may be secondary, which is attributable to an underlying cause, which may include<sup>3</sup> trauma to the joints, bony deformities, metabolic diseases, crystal deposition arthritides, fractures or loss of sensation to joints. Along with the above mentioned factors, the other risk factors for osteo-arthritis are age, female sex, race, genetic factors, repetitive stresses to the joints<sup>4</sup>.

It follows that malfunction of a joint may result from either acute or chronic injuries that produce bio-mechanical and biochemical forces leading to anatomic alterations in the shape of articulating surface and leading to matrix synthesis, enzymatic degradation of the connective tissue matrices resulting from an inflammatory conditions ( e.g. rheumatoid arthritis)

Moderate macroscopic and histological evidence of synovial inflammation was found in 55% of patients with knee osteo-arthritis and normal radiographs by arthroscopy<sup>5</sup>. Modest increases of the sensitive acute-phase proteins SAA and CRP have been observed in several studies of OA as well as in post-traumatic joint disease<sup>6,7</sup>. It may be argued that the chondrocyte in OA releases mediators very similar to activated macrophages. Cytokines and growth factors like interleukin-1B, tumour

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necrosis factor-B, TPA (tissue plasminogen inhibitor) may function to activate enzymes involved in proteolytic digestion of cartilage. These enzymes, a family of matrix metalloproteins (MMPs), including stromelysin, collagenase and gelatinase can degrade all the components of the extracellular matrix at neutral pH / 10<sup>4</sup>. TIMP (tissue inhibitor of metalloproteinase) and PAI-1 (plasminogen activator inhibitor) limit the degradative activity of the MMPs and plasminogen activators.

Diacerein is a pro drug which gives the active metabolite rhein. Rhein can effectively inhibit the IL-1 activated MAPK pathway and the binding of NF- $\kappa$ B and AP-1 transcription factors, two key factors involved in the expression of several pro-inflammatory genes by chondrocyte 8, 9-52, 53. In addition the drug can reduce the pro-catabolic effect of the cytokine, by reducing the MMP-1 synthesis and enhance the synthesis of matrix components such as type II collagen and aggrecan<sup>8,9</sup>. It increases the production of transforming growth factor- $\beta$  (TGF- $\beta$ 1 and TGF- $\beta$ 2) that stimulates chondrocyte proliferation and stimulates the production of collagen II, proteoglycan synthesis and increase matrix components.

The pharmacokinetic behaviour of diacerein is the same in young and elderly people with normal renal function after a single dose (50 mg) or repeated doses (25 to 75mg) twice daily. Taking diacerein with a standard meal delays systematic absorption but is associated with a 25% increase in the amount absorbed. Hence it is advisable to administer the drug with a major meal. Though dose modification is required in mild to severe renal insufficiency (50% reduction in severe renal failure) tab<sup>10</sup>, no reduction in initial dose is proposed in liver cirrhosis tab<sup>11</sup>.

Two adverse effects of diacerein are important to note. The first is a laxative effect, associated with diarrhoea or soft stools in 20% to 30% of patients after the first few doses whose frequency decreases with the continuation of treatment. The second adverse effect is yellow-brown discolouration for which the patient needs to be advised to prevent unnecessarily feeling of fear or anxiety.

Table 1 gives an analysis of efficacy and adverse effects of diacerein in the treatment of osteo-arthritis of knee as published by the various authors<sup>12-15</sup>.

### Materials and Methods:

A prospective, randomised, controlled clinical study was carried out in the Department of Physical Medicine and

Rehabilitation, AIIMS, New Delhi. A total of 76 patients were included in the study, divided into two groups, A and B, 38 patients in group A given diacerein and aceclofenac and 38 patients in group B only aceclofenac in predetermined doses. All the patients of both groups were given home bound strengthening exercises for the quadriceps and hamstrings muscles in the following protocol-straight leg rise, short arc extensions (knee), 30 repetitions, initially with ½ kg weight for resistance and gradually increased by ½ kg for every 4 weeks up to 2 kg in leg (B/L), 6 seconds holding time in each, twice in a day, to continue throughout the study. Physical modalities were also advised to reduce pain.

In group A, the patients were given diacerein 50 mg once daily for 4 weeks, then 50 mg twice daily for the next 2 months. Along with it, they were given aceclofenac 100 mg twice daily for 1 week then continued aceclofenac 100mg on SOS basis. In the group B, the patients were given aceclofenac 100 mg twice daily for 1 week then continued aceclofenac 100 mg on SOS basis.

Each patient was given a questionnaire page to answer the assessment questions, according to their subjective feelings about pain, stiffness and difficulties in functional activities using VAS and WOMAC scale at the initiation of study, at the 6th week and finally at the 12th week. Along with it, the frequency of intake of aceclofenac was monitored in the two groups. At the end of three months, the final assessment was taken and the desired study results were analysed using SPSS software.

### Assessment:

The symptoms and functional disabilities on performance of activities of daily living of the two groups of patients were assessed using The Western Ontario and McMaster Universities (WOMAC) osteo-arthritis index. The Western Ontario and McMaster Universities (WOMAC) osteo-arthritis (OA) index is a tested questionnaire to assess symptoms and physical functional disability in patients with OA of the knee and the hip. The WOMAC is a patient-centred, self-reported measures which provides opportunity to evaluate consequences of osteo-arthritis, that are important and relevant to patients with the condition<sup>16,17</sup>. The questionnaire contains 24 questions, targeting areas of pain, stiffness and physical function, and can be completed in less than 5 minutes. The self-rating instrument consisting of 24 items of which 5(P1-P5) relate to pain, (S1-S2) to joint stiffness, and 17(F1-F17) to the function scale. All items are rated

on a numerical rating scale of 0(no symptoms/disability) to 10(maximal symptoms/ disability).The unweighted arithmetic mean of at least 4/5 pain, 1/2 stiffness and 14/ 17 disability items make up the WOMAC scales.

**Visual analogue scale (VAS):**<sup>18</sup> The VAS is a self-report instrument that consists of a 10 cm straight line

of either horizontal or vertical orientation. The line is anchored by two extremes of pain. “no pain” and pain as bad as it could be. Possible score ranges from 0 to 10. The resulting measure represents the patient’s level of pain.

Score: 1-4=mild, 5-6=moderate pain, 7-10=severe pain.

**Table 1:** Efficacy and Adverse Effects of Diacerein in Various Studies

Authors with study	Efficacy, dosage recommendations	Safety profile and recommendation
Bernhard Rintelin <i>et al</i> <sup>12</sup> in their meta-analysis of 19 studies	Diacerein was significantly superior to placebo during the active treatment phase (Glassgow score, 1.50 [95% confidence interval, 0.80-2.20]. Diacerein, showed a carryover effect, persisting up to 3 months after treatment, with a significant analgesic-sparing effect during the follow-up period (Glassgow score, 2.06 [95% confidence interval, 0.66-3.46])	Tolerability assessment revealed no differences between diacerein and NSAIDs, although the latter showed more severe events
Zheng <i>et al</i> <sup>13</sup> on a study with 223 patients satisfying the ACR criteria for OA knee, to look for the efficacy rates with patient’s/physician’s overall assessment on diclofenac alone versus diclofenac with diacerein after 12 weeks of treatment.	Consumption of paracetamol was significantly lower in diacerein group than in the diclofenac group during the follow-up	Adverse events were 35.7% in diacerein and 45.1 % in the diclofenac group
Pelletier <i>et al</i> <sup>14</sup> Randomised, double-blind, placebo-controlled,, parallel study group with 3 diacerein dosages of 50 mg/day, 100 mg/day and 150 mg/day (administered twice daily).	Diacerein dosage of 100 mg/day was significantly superior (P<0.05) to placebo using the primary criterion (VAS assessment of pain) and the secondary criteria, which included the Western Ontario and McMaster Universities OA Index (WOMAC), and the VAS assessment of handicap	The best daily dosage of diacerein was 90.1 mg. Mild-to-moderate transient changes in bowel habits were the most frequent adverse effects, increasing with the dosage
Brahmachari <i>et al</i> <sup>15</sup> . Efficacy and safety of diacerein in early knee osteoarthritis:a randomised placebo-controlled trial	Diacerein showed highly significant (P<0.01) reductions in VAS pain scores, significant in WOMAC physical functional scores, significant lower requirements for rescue medications	Incidence of AE was significantly higher in diacerein arm(27 patients on diacerein experienced AE versus eight with placebo; p<0.01) with yellow discolouration of urine, soft stool, dyspepsia, mild to moderate intensity abdominal cramp, and skin rash being most common events
Pavelka <i>et al</i> <sup>2</sup> . A randomised, multicenter, double-blind, placebo-controlled study on 168 patients with primary end points at two months after the end of a three-month treatment period	At month 5, diacerein showed statistically significant superiority versus placebo as assessed with both the WOMAC A (P <0.0001) and the total WOMAC (P<0.0001), demonstrating the carryover effect of the drug. This superiority was already evident from month 2 for pain (P =0.001) and month 1 for total WOMAC (P _ 0.0021)	Diacerein was safe and well tolerated. No serious orpreviously undocumented adverse events were observed during the study

## Results and Observations:

### Demographic and clinical variables of study subjects:

The age at presentation of patients in group A varied from 41 to 68 years with mean age of 54.57% and in group B from 41 years to 72 years with mean age of 53.06%. The maximum number of patients was in the age group of 50 to 59 years. Female patients were more than male in both the groups, 22 females (62%) in group A.

**Progression of the disease:** We had found that 20%, 51.42%, 22.85%, 5.71 % respectively of the patients in group A had disease progression measured as stages I, II, III and IV according to the Kellgren-Lawrence staging of knee OA. The corresponding figures in the group B were 22.22 %, 58.33%, 22.22% and 5.81% respectively. After comparison of all history of both the groups, we found there was statistically no significant difference between the two groups at the inclusion of the study.

**Duration of the disease:** We had found that the average duration of disease in group A was 38.29 months and 39.39 months in group B. Maximum number of patients were those who have been having the disease for 2-3 years in both the groups, 9 (25.71%) in group A and, 10 (27.77%) in group B. There was no statistically significant difference between the groups in relation to the duration of knee pain ( $p=0.666$ ).

**Joint compartments involvement:** There were 48.50% of patients in group A and 44.44% patients in group B had bilateral compartments involvement, whereas only

17.14% and 27.77% respectively of patients in group A and group B had unilateral compartment involvement, remaining 34.25% in group A and 27.72% in group B had involvement of all the compartments of the knee. They were compatible at both groups. There was no statistically significant difference between the groups,  $p=1.0$

**Intensity of knee pain and stiffness:** Table 2 shows the changes in the various sub-scores over the period of time. The mean VAS score of pain on the sub-score (pain while walking) were 4.80, 3.06 and 1.80 at 0, 6th and 12th weeks respectively for patients in the group A which was statistically significant ( $p=0.00$ ). The findings of the other sub-scores are also shown in the table. This was reduced over the period of time and was highly significant at the completion of the study ( $p=0.000$ ). At the end of the study, the p-value was 0.000 which was also statistically significant between both the groups.

**Stiffness of knee joint:** In Table 3, where we compared the numerical rating scale (0-10) in stiffness at early morning time and stiffness later at the day within the group and between the groups at inclusion 0 week, 6th week and at the completion of the study. They were comparable within the groups and in both group A and group B at the starting of the study. And in between the groups, they were comparable but after 6 weeks of time ( $p=0.000$ ). Here it was found significant difference between both the groups ( $p=0.00$ ). The improvement was better with group A over group B at the end of the study.

In Table 2, we have compared the difficulty level in

**Table 2:** Mean Vas Score of Pain

Variable	Group	0 week	After 6 weeks	After 12 weeks	p-value
		Mean VAS score	Mean VAS score	Mean VAS score	
P1 Pain while walking	A	4.80	3.06	1.80	0.000
	B	4.78	3.33	2.56	0.000
P2 Climbing wtairs	A	6.86	4.77 1.087	2.94	0.000
	B	6.83	4.92	3.67	0.000
P3 In bed at night	A	1.83	1.40	1.09	0.000
	B	1.94	1.44	1.08	0.000
P4 Sitting or lying	A	2.00	1.46	1.11	0.000
	B	1.94	1.39	1.14	0.000
P5 Standing	A	5.69	3.89	2.29	0.000
	B	5.25	3.81	3.03	0.000

**Table 3:** Numerical Rating Scale

Variable	Group	0 week	After 6 weeks	After 12 weeks	p-value
		Mean RS	Mean RS	Mean RS	
S1	A	3.11	1.94	1.31	0.000
	B	2.78	1.81	1.39	0.000
S2	A	2.57	1.69	1.43	0.000
	B	2.86	2.00	1.83	0.000

carrying out the various activities of daily livings as according to the WOMAC scale. At the beginning of the study, the mean numerical rating scale (0-10) of the various parameters are compared within the groups and between the groups and was comparable in between the groups and among the groups. Over the course of time, the improvement was much better with group A than that with the group B in all the parameters,  $p=0.000$ .

**Reduction in the intake of NSAIDs:** The average number of NSAIDs (tablets) taken by the patients in the group A and group B and their gradual decrease over the period of twelve weeks after starting treatment with diacerein is shown in the Figs 1 and 2 and Tables 4 and 5. The reduction has been found to be statistically significant ( $p=0.000$ ) in both the groups. Comparing the reduction among the two groups, patients in the group A had lesser number of NSAIDs intake as compared to the group B as shown in the Figs 1 and 2.

One patient in group A dropped out due to allergic reaction to diacerein like skin rashes, itching. The symptoms subsided after stopping diacerein. Although soft stool, diarrhoea and mild abdominal discomfort occurred in few patients, none of the patients dropped out due to these symptoms. These symptoms also subsided within a few days even in continuing with the drug. Some had abdominal pain and discomfort which may be due to side-effects of aceclofenac, which was later treated with PPIs (proton pump inhibitors). The dropout were due to various reasons like missing the drug in between and few of them could not come on time mainly those who were from very far off places.

### Discussion:

Osteo-arthritis is one of the most frequent causes of pain, loss of function and disability in adults second to ischaemic heart disease. In India the prevalence of osteo-arthritis has been suggested at 24.9%<sup>19</sup> till recently, osteo-arthritis was classified as a wear and tear disorder

of articular cartilage for which only pain reducing medications, physiotherapy and other joint injections with corticosteroids were the mainstay of treatment. Very little scientific consideration has given on the modification of course of disease activity leading to musculoskeletal disability and affecting quality of life.

The present treatment of osteo-arthritis revolves around the use of analgesics and NSAIDs and newer cyclooxygenase-2 inhibitors. But these medications increases the risk of upper gastro-intestinal adverse effects and may have significant renal and cardiovascular toxicity especially NSAIDs including the COX-2 inhibitors<sup>20</sup>. Moreover, they don't have the disease course modifying effect. Recently, the used of symptomatic slow acting drugs or chondroprotective drugs like glucosamine, chondroitin sulphate and diacerein<sup>21</sup>. These drugs have gradual onset of action (4-6 weeks) but maintain their symptomatic effect for a period of 4-8 weeks after cessation of treatment.

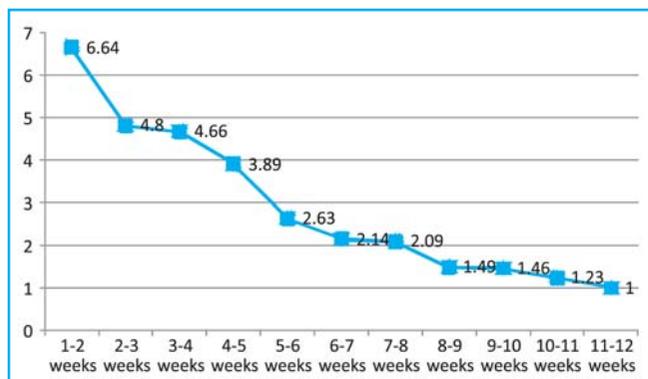
Diacerein acts by inhibiting the inflammatory mediators like IL-1, and MMP-1 and by enhancing the synthesis of matrix components such as type II collagen and aggrecan<sup>8,9</sup>. It increases the production of transforming growth factor-b (TGF-b1 and TGF-b2) that stimulates chondrocyte proliferation and stimulates the production of collagen II, proteoglycan synthesis and increases matrix components. Diacerein has been found to be safe and can be given to those with liver diseases but reduction of dose is needed to those with severe renal failure.

In our study, the group of patients (group A) who were given diacerein at the dose of 50 mg daily for one month to be followed by 50 mg twice daily for another two months were compared to the group of patients (group B) who were not given diacerein. Both groups of patients were prescribed a uniform exercise regime to be followed at home on a regular basis, joint care and protection advice given and physical modalities suitable to the

**Table 4:** Average Number of Aceclofenac 100mg Tablets Taken per Week in Group A

Weeks	Mean	Std. Deviation
A (1-2) weeks	6.46	1.221
B (2-3) weeks	4.80	1.079
C (3-4) weeks	4.66	0.968
D (4-5) weeks	3.89	0.993
E (5-6) weeks	2.63	1.165
F (6-7) weeks	2.14	1.141
G (7-8) weeks	2.09	1.197
H (8-9) weeks	1.49	0.853
I (9-10) weeks	1.46	0.950
J (10-11) weeks	1.23	0.973
K (11-12) weeks	1.00	0.840

**Fig 1-** Average Number of Aceclofenac 100mg Tablets Taken per Week in Group A



patients were also recommended. The mean VAS score of pain on the sub-score (pain while walking) were 4.80, 3.06 and 1.80 at 0, 6th and 12th weeks respectively for patients in the group A which was statistically significant ( $p=0.00$ ). Significant changes were seen on the other sub-scores of pain, stiffness and difficulty in functional activities included in the WOMAC scale. The effects of diacerein were observable by 6th week period and sustained at the end of the third month.

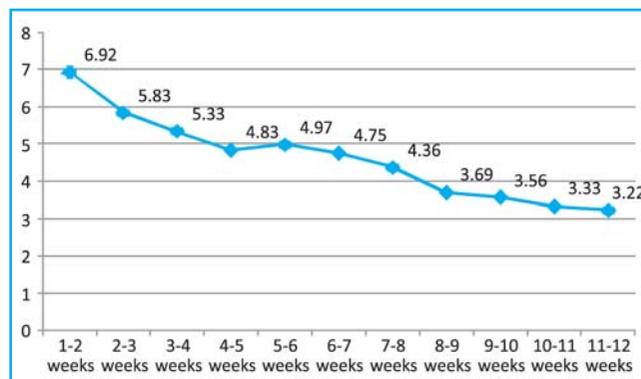
The average number of NSAIDs (aceclofenac 100 mg tablets) taken by the patients in the group A and group B has been found to be decreased and were statistically significant ( $p=0.000$ ) in both the groups. Comparing the reduction among the two groups, patients in the group A had lesser number of NSAIDs intake as compared to the group B.

One patient in the diacerein group dropped out due to skin rashes and itching which might be due to allergic reaction to diacerein. The symptoms subsided after

**Table 5:** Average Number of Aceclofenac 100mg Tablets Taken per Week in Group B

Weeks	Mean	Std. Deviation
A (1-2) weeks	6.92	1.251
B (2-3) weeks	5.83	1.082
C (3-4) weeks	5.33	1.195
D (4-5) weeks	4.83	1.000
E (5-6) weeks	4.97	1.028
F (6-7) weeks	4.75	0.874
G (7-8) weeks	4.36	0.990
H (8-9) weeks	3.69	0.920
I (9-10) weeks	3.56	0.969
J (10-11) weeks	3.33	0.535
K (11-12) weeks	3.22	0.797

**Fig 2-** Average Number of Aceclofenac 100mg Tablets Taken per Week in Group B



stopping diacerein. Soft stools, diarrhoea and mild abdominal discomfort occurred in few patients, these would subside within a few days, but none of the patients dropped out due to these symptoms. The dropout was due to various reasons like missing the drug in between and few of them could not come on time mainly those who were from very far off places.

### Summary and Conclusion:

Both the patients were statistically comparable, at the initiation of the interventions in relation to age, sex distribution, height, body mass index, occupations, socio-economic status, dietary and family history, duration of the knee pain, clinical features.

In both the patient groups, medications were not only helpful in improving the symptoms, but also activities of daily living of the patient with primary osteo-arthritis of knee. But those patients who were taking diacerein along with aceclofenac, pain, stiffness and activities of

daily living were much better with them than with aceclofenac alone. Most of the adverse effects of diacerein were mild and easily tolerable. To conclude, diacerein is an effective drug for osteo-arthritis of the knee with minimal side-effects and possible NSAIDs sparing effect which might be very helpful in preventing renal and gastric involvements in patients with a chronic disease like osteo-arthritis.

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## Editorial

### International Day of Persons with Disabilities

#### **“Break Barriers, Open Doors: For an Inclusive Society for All”**

United Nations’ (UN) International Day of Persons with Disabilities (IDPwD) is annually held on Dec 3 to focus on issues that affect people with disabilities. It re-affirms and draws attention to the rights of people who live with disabilities. From 1983 to 1992 UN observed “Decade of Disabled Persons” to improve the life of disabled. On October 1992 UN General Assembly proclaimed December 3 as the “International Day of Disabled Persons” and was first observe on 3rd December 1992. On 18th December 2007 the assembly change the name as International Day of Persons with Disabilities (IDPwD) and was first observe on 3rd December 2008.

The UN symbol consists of an azimuthally equidistant projection of the globe centred on the North Pole surrounded by olive branches. The word ‘enable’ is written in lower case, ‘e’ is read and the other letters are blue. Around 15% of the world’s population, or one billion people, live with disabilities. People are often unaware of the great number of the persons living with disabilities around the world and the challenges they face.

Persons with disabilities, “The world’s largest minority”, often face barriers to participate in all aspects of society. Barriers can take a variety of forms, including those relating to the physical environment or to information and communication technology (ICT), or those resulting from legislation or policy, or from societal attitudes or discrimination. The result is that persons with disabilities do not have equal access to society or services, including education, employment, health care, transportation, political participation or justice. The day aims to promote an understanding of the disability issues and mobilise support for the dignity, rights and well-being of persons with disabilities. It also seeks to increase awareness of gains to be derived from the inclusion of persons with disabilities in every aspect of life.

Earlier, the international disability movement achieved an extraordinary advance in 2006, with adoption of the Convention on the Rights of Persons with Disabilities. The convention is intended as a human rights instrument with an explicit, development dimension. On 23rd September 2011 UN General Assembly Convened the High Level Meeting on Development and Disability (HLMDD), under the theme “The way forward ; a disability inclusive development agenda towards 2015 and beyond”.

The meeting took place five years after UN Convention on Right of Persons with Disabilities entered into force, two years after released of the World Report on Disability and two years away from 2015 - the target date for the achievement of the Millennium Development Goals (MDGs).

It’s time to effectively implement the Outcome Document of the high level Meeting and to break barriers and open doors: to realise an inclusive society and development for all. International Day of Persons with Disabilities 2013 provides an opportunity to further raise awareness of disability and accessibility as a cross cutting development issue and further the global efforts to promote accessibility, remove all types of barriers, and to realise the full and equal participation of Persons with Disabilities in society and shape the future of development for all.

– R. N. Haldar

# Electrodiagnosis of Quadrilateral Space Syndrome: A Case Report

Dholakia Madhuri<sup>1</sup>, Sinha Anupam<sup>2</sup>

### Abstract

Quadrilateral space syndrome is a rare occurrence of compression of the axillary nerve within the quadrilateral space. It presents as shoulder pain with weakness due to teres minor atrophy. It can often times be overlooked and undertreated. MRI and electrodiagnostic studies are used to confirm the diagnosis. We present a case of a patient with quadrilateral space syndrome confirmed using both MRI and electrodiagnosis.

**Keywords:** Electrodiagnosis, quadrilateral space syndrome, axillary nerve.

### Case Report:

A 56 year-old male presented to our practice with a two year history of non-traumatic neck and right shoulder pain. He denied any upper extremity radicular pain but did report mild paresthesias in his hands. He denied any bowel, bladder, or balance disturbance. On examination, the patient was neurologically intact without evidence of upper motor neuron signs. He did have mild weakness in right shoulder strength along with positive shoulder impingement signs. MRI of the cervical spine showed evidence of C6-7 foraminal stenosis. The patient had undergone physical therapy and cervical epidural injections with marginal improvement of his symptoms. MRI of the right shoulder revealed mild supraspinatus tearing and fatty atrophy of the teres minor.

Electrodiagnostic testing of the right upper extremity showed normal nerve conduction and needle studies,

except for denervation found only in the teres minor; there was no denervation noted in the cervical paraspinals or remainder of the right upper extremity. The patient was diagnosed with quadrilateral space syndrome (QSS) and referred for physical therapy.

### Discussion:

QSS results from compression of the distal branch of the axillary nerve and/or posterior humeral circumflex artery as it exits this anatomic compartment<sup>1</sup>. Symptoms result from compression of the axillary nerve, not from arterial occlusion. The quadrilateral space is defined by the long head of the triceps (medially), surgical neck of the humerus (laterally), teres minor (superiorly), and teres major (inferiorly)<sup>2</sup> (Fig 1). QSS may often present secondary to anterior shoulder dislocation, impingement from a cystic mass, muscular hypertrophy, fibrous bands, gunshot wounds, iatrogenic injury during shoulder surgery, sports injury (most often in overhead athletes), and venous dilation<sup>3</sup>.

Diagnosis of QSS requires a high index of suspicion from the physician. Examination will reveal pain and/or weakness with shoulder abduction and external rotation. MRI of the shoulder will show isolated atrophy of the teres minor, often without findings in the deltoid. Subclavian arteriography and Doppler ultrasound are useful in diagnosing arterial occlusion<sup>4,5</sup>. Electrodiagnostic studies (EMG) may show denervation of the teres minor with possible findings in the deltoid as well.

Isolating the teres minor during EMG involves the patient lying on the unaffected side. The needle is inserted one

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third of the way between the acromion and inferior angle of scapula along the lateral border. The muscle is activated by externally rotating the shoulder<sup>6</sup>. Needle examination of the supraspinatus and infraspinatus muscles should be normal.

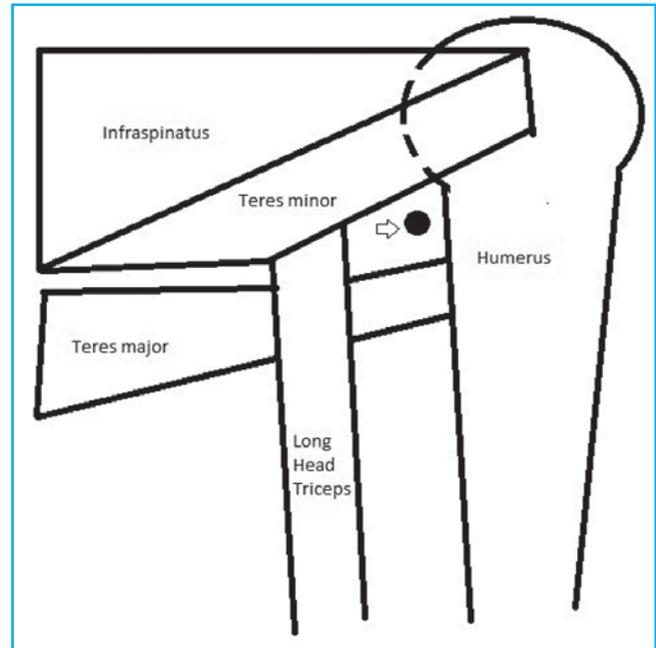
Treatment of QSS initially involves rest and non-steroidal anti-inflammatories, followed by a course of physical therapy. For refractory cases, surgical decompression of the axillary nerve may be considered as well<sup>3,5</sup>.

### Conclusion:

We present a rare case of right shoulder pain and weakness secondary to isolated teres minor atrophy from axillary nerve injury within the quadrilateral space. Quadrilateral space syndrome is often overlooked as a diagnosis in patients presenting with shoulder pain and weakness. Clinicians should be aware of this syndrome, especially in the young athlete, and should rely on both MRI and EMG of the shoulder to confirm the diagnosis.

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**Fig 1-** Arrow Pointing to the Axillary Nerve within the Quadrilateral Space



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## Intra-articular Steroid in the Management of Adhesive Capsulitis of Shoulder: A Comparison of the Anterior and Posterior Approaches

Sharma G Sonachand<sup>1</sup>, Nandabir Singh Y<sup>2</sup>  
Bimol Singh N<sup>3</sup>, Touthang Alex T<sup>4</sup>, Tamphaleima KH<sup>5</sup>

### Abstract

**Objective:** To compare the anterior and posterior blind intra-articular steroid injections approaches in the management of adhesive capsulitis of shoulder

**Design:** Randomised control trial

**Setting:** Department of PMR, Regional Institute of Medical Sciences (RIMS), Imphal, Manipur.

**Participants:** The patients having adhesive capsulitis of shoulder (n=60) attending PMR department, RIMS during the study period.

**Duration:** One year (August 2011 to July 2012).

**Intervention:** After randomisation, 60 patients were allocated in three groups (A, B and C). Group C (n=20) received physical therapy practice in the department of PMR, RIMS, Imphal. Group A (n=18) and B (n=22) received intra-articular steroid (methylprednisolone 80mg each) by anterior and posterior approaches without imaging guidance respectively in addition to physical therapy. Outcome measures: 1) Visual analogue scale (VAS) for pain, 2) Shoulder pain and disability index (SPADI) and 3) Passive range of motion of affected shoulder using goniometer.

**Results:** All the three groups showed improvement with statistically significant ( $p < 0.005$ ) findings in all the outcome variables except in shoulder flexion range ( $p = 0.085$ ). Improvement in outcomes namely VAS, SPADI, shoulder rotation and abduction range were more marked in group A when compared with group B which was found statistically significant ( $p < 0.05$ ).

**Conclusion:** The intra-articular steroid injection when combined with physical therapy is effective in the management of adhesive capsulitis of shoulder. The blind anterior approach intra-articular steroid injection is more effective than posterior approach in improving shoulder rotation and abduction range of movements, reducing shoulder pain and disability in patient having less than 3 months duration adhesive capsulitis of shoulder.

**Key words:** Gutter crutch, adhesive capsulitis, intra-articular steroid, visual analogue scale, shoulder pain and disability index, passive range of motion.

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### Introduction:

Adhesive capsulitis is one of the common causes for shoulder dysfunction. It is defined as “a painful restriction in shoulder range of motion in a patient with normal radiograph”<sup>1</sup>. It is usually an idiopathic self limiting condition, but it can be associated with other secondary pathological conditions<sup>2</sup>. The loss of passive range of movement, particularly related to external rotation, has remained pivotal to the diagnosis of adhesive capsulitis of shoulder. The treatment options documented in the literature include supervised physical rehabilitation<sup>3-6</sup>, non-steroidal anti-inflammatory medications<sup>7,8</sup>, oral corticosteroid<sup>9</sup>, intra-articular corticosteroid injection<sup>4,10-12</sup>, distension arthrography<sup>13</sup>,

closed manipulation<sup>13-17</sup>, open surgical release, and arthroscopic capsular release<sup>17-20</sup>.

Intra-articular (glenohumeral joint) steroid injection is effective during early stage of the adhesive capsulitis. The rationale for corticosteroid injection is to reduce synovial inflammation, decrease capsular fibrosis and allow improvement of motion with a decreased time to functional recovery<sup>21</sup>. The intra-articular injection of shoulder joint can be performed either anterior (Fig 1) or posterior approaches (Fig 2) with or without imaging guidance. In our state, because of its ease and low cost, the blind glenohumeral injection is performed commonly for the management of adhesive capsulitis of shoulder. We hypothesised that the anterior approach intra-articular steroid injection is more effective than posterior one in the management of early stage adhesive capsulitis of shoulder.

### Aims and Objects:

To see the effectiveness of intra-articular steroid in the management of adhesive capsulitis of shoulder.

To compare the effects of anterior and posterior approaches blind intra-articular steroid injection in early stage adhesive capsulitis of shoulder.

### Materials and Methods:

It is a randomised controlled trial. Setting conducted in the Department of Physical Medicine and Rehabilitation (PMR), Regional Institute of Medical Sciences (RIMS), Imphal, Manipur with study period of 1 year (August 2011 to July 2012). All the patients having adhesive capsulitis of shoulder attending PMR department, RIMS, during the study period and fulfilling the inclusion criteria were included in the sample size.

#### Inclusion criteria:

1. Patient having unilateral adhesive capsulitis of shoulder with duration less than 3 months.
2. Not received any intra-articular injection earlier.
3. Age between 35 and 70 years.

#### Exclusion criteria:

1. Patient unable to or not willing to give consent. Bilateral involvement.
2. Non-cooperative patients.
3. Recent trauma or fracture around the shoulder.
4. Recent history of any operative interventions.
5. Having comorbid conditions like uncontrolled diabetes, chronic renal failure, thyroid diseases, coronary artery disease, stroke, connective tissue disorders etc.

**Study groups:** Altogether 60 patients attended during the study period. Subjects (n=60) were randomly allocated into three groups namely A(n=18), B(n=22) and C(n=20) by using simple randomisation by lottery method.

### Interventions:

All the groups received physical therapy for adhesive capsulitis practised in the department of PMR, RIMS, Imphal. It included 1) Passive range of motion exercise, 2) moist heat therapy and 3) stretching exercises. The above physical therapy was given 10 sessions per day for 4 weeks. Groups A and B received intra-articular methylprednisolone (depomedrol) 80mg by blind method in addition to physical therapy. Drug was deposited in the shoulder joint through anterior approach in group A and posterior approach in group B under strict aseptic precautions. The anterior approach involved inserting the needle tip a fingerbreadth lateral to and just below the coracoid process and directing the needle posteriorly and slightly laterally while the patient was in supine with arm externally rotated<sup>22,23</sup>. For the posterior approach, the patient was put in sitting position and the needle was inserted two fingerbreadths inferior and medial to the posterolateral corner of the acromion and directed anteromedially towards the coracoid process<sup>24-27</sup>.

#### Groupwise intervention:

- Group A - Intra-articular steroid (methylprednisolone 80mg) shoulder joint, anterior approach and physical therapy.
- Group B - Intra-articular steroid (methylprednisolone 80mg) shoulder joint, posterior approach and physical therapy.
- Group C - Physical therapy.

**Follow-up:** Follow-up was done 1st at one week and lastly at one month.

**Outcome measures:** The following outcome measures were taken before intervention and during subsequent follow-up;

1. Visual analogue scale (VAS) for pain
2. Shoulder pain and disability index (SPDI)
3. Passive range of motion of affected shoulder using goniometer.

**Data analysis:** ANOVA was used for the comparison between the groups. A significant level of 0.05 was used for all comparisons.

**Ethics:** All the participants were informed about of the nature of the study and those agreed to participate were asked to sign the informed consent form.



**Fig 1-** Anterior Approach



**Fig 2-** Posterior Approach

Participants were assured that they could withdraw from the project at any time. The approval of the institutional ethics committee, RIMS, Imphal, Manipur, was taken.

**Results:**

Sixty subjects (45% male, 55% female) were recruited and randomly assigned to three groups. Eighteen subjects were enrolled in group A (steroid injection; anterior approach and physical therapy), 22 in group B (steroid injection; posterior approach and physical therapy), and 20 in group C (physical therapy). Three subjects did not return for all follow-up visits; two in group B and one in group C.

No disparity was found between the baseline characteristics of each group including age, duration of symptoms, shoulder range of motion, VAS and SPADI (Table 1). The non-dominant side was affected in less than half of the cases ( $n = 60$ ; 40%). Compliance with physiotherapy was good with 90% completing all sessions in study period. At 1st follow-up, there were

improvement in all the outcome measures in all the three groups and statistically significant ( $p < 0.005$ ) improvement were found for VAS, SPADI and abduction shoulder range (Table 2). At last follow-up, there was improvement in all the three groups with statistically significant finding ( $p < 0.005$ ) in all the outcome variables except in shoulder flexion range ( $p = 0.085$ ). Improvement in outcomes namely VAS, SPADI, shoulder rotation range were more marked in group A when compared with group B and differences were found significant statistically ( $p < 0.05$ ), as evidenced by ANOVA Post Hoc Test (Tables 3 and 4).

**Discussion:**

The present study showed that improvement in overall shoulder disability and pain both at 1 week and 1 month which were better in subjects treated with corticosteroid injection and physical therapy than those treated with physical therapy alone. These results support the findings of previous studies suggesting improvement in early outcome after corticosteroid injection in adhesive

**Table 1:** Baseline Clinical Characteristics among the Groups

Variables	Group A (Mean ± SD)	Group-B (Mean ± SD)	Group-C (Mean ± SD)	F value	p-value	
Age	52.00 ± 1.26	51.90 ± 9.00	46.00 ± 6.90 SD	2.851	0.066	
Duration	1.70 ± 0.80	1.70 ± 0.55	2.20 ± 0.70 SD	3.270	0.142	
VAS	6.20 ± 1.20	6.50 ± 1.05	6.40 ± 1.00	0.212	0.809	
SPADI	59.70 ± 8.70	64.50 ± 8.90	85.80 ± 9.80	1.150	0.321	
Passive ROM	Abduction	88.80 ± 2.30	98.10 ± 2.70	90.50 ± 2.70	0.735	0.484
	Flexion	105.28 ± 1.80	112.20 ± 2.30	107.20 ± 2.30	0.533	0.589
	External rotation	32.20 ± 1.90	30.60 ± 1.60	27.20 ± 2.00	0.359	0.700
	Internal rotation	27.70 ± 2.00	33.80 ± 2.20	34.00 ± 2.10	0.500	0.609

**Table 2:** Difference in Outcomes between the Groups at 1 Week

Variables		Group A (Mean ± SD)	Group-B (Mean ± SD)	Group-C (Mean ± SD)	F value	p-value
VAS		0.94 ± 0.63	3.20 ± 1.70	3.90 ± 0.82	30.71	0.000
SPADI		23.20 ± 2.08	34.30 ± 1.20	54.90 ± 4.62	48.60	0.000
Passive ROM	Abduction	141.30 ± 39.80	124.50 ± 27.60	69.20 ± 40.10	21.45	0.000
	Flexion	152.70 ± 2.90	130.40 ± 2.60	124.70 ± 2.50	5.490	0.007
	External rotation	57.50 ± 2.60	39.50 ± 1.80	48.15 ± 1.40	3.976	0.024
	Internal rotation	61.10 ± 2.70	45.00 ± 1.90	45.70 ± 9.90	4.060	0.022

**Table 3:** Difference in Outcomes between the Groups at 1 Month

Variables		Group A (Mean ± SD)	Group-B (Mean ± SD)	Group-C (Mean ± SD)	F value	p-value
VAS		0.55 ± 0.50	1.80 ± 1.30	2.75 ± 1.20	18.50	0.000
SPADI		1.80 ± 2.70	14.10 ± 8.60	28.30 ± 9.60	54.76	0.000
Passive ROM	Abduction	174.40 ± 1.90	148.60 ± 2.60	77.50 ± 3.90	54.72	0.000
	Flexion	174.40 ± 1.50	137.70 ± 3.70	159.10 ± 2.30	2.57	0.085
	External rotation	89.40 ± 2.30	56.10 ± 2.10	40.90 ± 1.10	52.22	0.000
	Internal rotation	69.40 ± 2.40	60.00 ± 1.90	39.00 ± 1.40	57.91	0.000

**Table 4:** Comparison in Outcomes between Group A and B at 1 Month

Variables Between Group A & B	Mean Difference (A - B)	Standard Error	Significant	95% confidence interval	
				Lower value	Upper value
VAS	-1.308	0.354	0.001	-2.160	-0.455
SPADI	-12.247	2.475	0.000	-18.205	-6.290
Flexion	-53.282	72.385	0.743	-227.472	120.906
Abduction	25.808	9.504	0.023	2.935	48.680
External rotation	33.308	4.729	0.000	21.928	44.688
Internal rotation	29.444	4.591	0.000	18.395	40.493

capsulitis capsulitis of shoulder<sup>28,29</sup>. Reasons for choosing methylprednisolone (depomedrol) in our study are it is intermediate acting partially soluble steroid having sustained anti-inflammatory action because of its depot form, has very minimal mineral corticoid action and less side-effect.

The improvement in the shoulder range of motions (rotation and abduction) and pain was significantly better at 1 month in subjects receiving physical therapy and intra-articular steroid by anterior approach when compared with those receiving physical therapy with intrarticular steroid by posterior approach. The rotation (external and internal) and abduction motions are thought to be the first movements to be affected in the adhesive capsulitis of shoulder<sup>30</sup>. The possible pathology may be due to adhesion of dependent part of inferior capsule,

contracture of coracohumeral ligament and anterior capsule. Direct deposition of sustained acting steroid in and around the anterior capsule may help in prevention of early pathological process occurring in adhesive capsulitis of shoulder. A cadaveric study conducted by Paul *et al*<sup>31</sup> showed that the anterior glenohumeral injection is 80% likely to be accurately placed in contrast to a 50% accuracy rate when the injection is placed posteriorly. Another study<sup>32</sup> on ultrasound guided suggested that the needle placement using the anterior technique was accurate in 96% of cases while that of the posterior technique was only accurate in 41%. So, the needle placement in the glenohumeral joint using anterior approach is more accurate than the posterior approach. This can also be the possibility why the anterior approach gives better outcome results in the present study.

## Conclusion:

The intra-articular steroid injection when combined with physical therapy is effective in reducing shoulder disability and pain in patient having 3 months duration adhesive capsulitis of shoulder. The anterior approach blind intra-articular steroid injection is more effective than posterior approach in improving shoulder range of motion (rotation and abduction), reducing shoulder pain and disability in early stage adhesive capsulitis of shoulder.

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## Obituary

### Dr Krishna Kumar Singh



You could see him in every IAPMR conference invariably dressed in a brown over coat supporting four large pockets with a belt, hanging, not tied; conferences are generally held during the beginning of the year when there is a bit of nip in the air. Many would have heard him point to the health budget of India being only 1.5 paisa per person per annum in those days and compare it with the world figures quoting WHO and the developed countries. Everyone was impressed with the figures he quoted. He would have a comment and occasionally a question from almost all the speakers on the dais. With his shiny silvery white hair well groomed wearing the heavenly 'teeka' from the visit to the temple earlier during the day, he would outshine all those around. With his slightly beetle stained full smile he would befriend anyone, young or old. His habit of meeting everyone even if for a short period attracted many towards him who did not know him and those who knew him would crowd around him to exchange a note of laughter or a serious matter, let it be the amendment of the RCI Act or the matter of the persons with disability with the ministry or the curriculum modification with the MCI, he would be head over heels to have his say in the matter through the association or with the government's machinery. He would use his resources to pool knowledge or to have it conveyed appropriately to the proper authority he would not stop. He himself was an authority and knew how to execute that. Hailing from the holy city of Patna, he had his spell all over the country, being the Honorary Surgeon to the President of India is not a small deal, Dr Krishna Kumar Singh, popularly known as 'KK' was one of the heaviest and the most emotionally charged President of our IAPMR during whose tenure we had our first international conference at AIIMS in 1991. Dr SK Varma organized the conference and the younger lot including me was one of the runners to execute the orders. Dr KK Singh's presence was felt all through the organizing two years by all of us. Seeing how people worked, he would come in and order 'tea and samosa' from our canteens and treat all. It was the first time the international conference was being held and the stakes were high with IAPMR with the proud feeling by the president and the organizers, he did not like to take the whole credit to himself. It was the first time he initiated the 'scroll of honor' awarded to the core workers in the conference organizing team, I as one of them could see the whole team was happy to the depth of one's heart due to this recognition given during the valedictory function of the conference. This was the style, manner, empathy and zeal of Dr KK Singh. Having had his innings at Patna Medical College he joined AIIMS and was here for almost eight happening years. He changed the face of the department and the style of working with his innovative methods. It was the first time the head of the department in the institute did not go to the administrator but asked him to come and meet him in the office of the head, such was his authority. The office of the department instead of having one PA all these years was teeming with activity with a full brigade of five. It was famous all over the institute that one must visit the office of the head of the department of PMR which had an entry two rooms away with a carpeted floor and two ante-rooms and a conference room before one could have the privilege of seeing the head of the department sitting pretty besides the 'water falls' in the room ready with a thermos flask full of tea specially brought from his home tightly brewed. No one could leave un-impressed. Dr KK Singh had a vision to change the face of PMR and he was all through his years inclined to work towards that. Unfortunately, his vision was not paralleled by his associates but that did not deter him to do what he wanted to including making PMR department at AIIMS to be recognized on the map of WHO. After he finished his term at AIIMS in 1998 he did not remain in the active life of the PMR or its association. It appeared that he had done what he wanted to and left youngsters to take the lead. Perhaps his health did not allow him to continue follow his dream but he did show the path to the gen-next. His journey to the heaven started on October 3, 2013 with a peaceful last moments with his son by his side asking him to give him a blanket since he was feeling cold. He just took a turn in the bed to take a bigger turn from this earth to the heavenly abode. He lived King style and ended his journey not less than an Emperor with his two good sons and a lovely daughter and a legacy for the PMR, unforgiveable. Our hearts dole out for the 'hero' of PMR.

Dr U Singh

### Erythema Multiforme in a Case of Polyarthritis

Pramanik R<sup>1</sup>, Das P<sup>2</sup>

A thirty five years female patient presented in PMR OPD with polyarthritis including small joints with morning stiffness for approximately 45 minutes for last two months. She was seropositive (positive RF and anti CCP) without any significant deformity. Her ESR and CRP were significantly high. We started the pharmacological treatment with methotrexate,



**Fig 1-** Showing Lesions over Leg

sulphasalazine, hydroxychloroquine and etoricoxib. She was also advised for energy conservation and joint protection technique, standardised exercise schedule, contrast bath etc.

She was feeling much better with reduction of DAS score in her next visit after 6 weeks and managing her job quite well. Suddenly she came back again after two weeks with macular lesions spreading centrifugally over her both legs. She didn't suffer from any recent sore throat, mucosal lesion, fever or malaise. On examination the lesions (Fig 1) were looking like classical target lesion i.e. erythematous macular spreading lesions with few central blisters over antero-medial surface of her both legs. We didn't find any herpetic lesion anywhere in her body. We along with our Dermatologist concluded this rash as drug induced erythema multiforme in patient of rheumatoid arthritis.

Then sulphasalazine was stopped and the skin lesions disappeared within next five days. Now the lady is doing well on methylprednisolone, methotrexate, and hydroxychloroquine with normal FIM score.

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# Plexiform Neurofibromatosis with Abnormal Pachydermatocele: A Rare Case Report

Agarwal AK<sup>1</sup>, Gupta Anil Kumar<sup>2</sup>, Kumar Dileep<sup>3</sup>, Kumar Vijay<sup>4</sup>, Yadav Ganesh<sup>5</sup>

### Abstract

Plexiform neurofibromatosis, also known as pachydermatocele, is a rare type of neurofibromatosis, caused by excessive growth of the neural tissue in the subcutaneous fat. This condition is rare in Indian subcontinent but when it occurs, it causes cosmetic as well as functional disability. The optimal management of plexiform neurofibroma is not well defined and surgery is often delayed. We report a rare case of neurofibromatosis with pachydermatocele involving the nerves of the left thigh and leg with disfigurement and associated disability.

**Keywords:** Plexiform, neurofibromatosis, disability.

### Introduction:

Neurofibromatosis types 1 and 2 (NF1, NF2) are autosomal dominant disorders that primarily affect the development and growth of nerve cell tissues<sup>1</sup>. Plexiform neurofibroma, also known as pachydermatocele, is a type of neurofibromatosis, caused by excessive growth of the neural tissue in the subcutaneous fat. It is also seen in connection with the branches of trigeminal nerve<sup>2</sup>. It has also been reported in retroperitoneal region, paraspinal and mediastinal area and anterior abdominal wall<sup>3,4</sup>. Plexiform neurofibroma is reported to occur in 26.7% of patients with type I neurofibromatosis<sup>5</sup>. The optimal management of plexiform neurofibroma is not well defined and surgery is often delayed until significant disfigurement has

occurred. This condition is commonly seen in the west, but rarely found in the Indian subcontinent<sup>6</sup>. We report a rare case of neurofibromatosis with pachydermatocele involving the nerves of the left thigh and leg with disfigurement and associated disability.

### Case Report:

We have admitted an adolescent of 16 years with the chief complaint of hand on knee gait on left side at the Department of Physical Medicine and Rehabilitation, C.S.M. Medical University, Lucknow. He also gave the history of minor injury to left knee region followed by effusion which was aspirated by a local physician followed by POP immobilisation one year back. At the time of the injury, x-ray of the patient was within normal limit. One year back, he had apparently no knee joint problem. On detailed interrogation of this patient, he also complained of hypertrophy of left thigh and upper third of the left leg with very soft, flabby, elastic swelling in the skin fold (Fig 1) since childhood. His parents informed that at the time of birth, he had hyperpigmented patches on anterior abdominal wall with no other skin or skeletal abnormality. His milestones were normal and became ambulatory within 14 months of birth.

On examination of the case, we found apparent lengthening of left lower limb, nearly 15 hyperpigmented patches on whole of the body (Fig 2). The size variation was 1cmx1cm to 3.5cm × 2cm. The less than 1.5cm patches were 8 and more than 1.5cm patches were 10 in number. He has painless, extra fold of skin which is soft,

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mobile, without resistance and tenderness giving a picture of pachydermatocele. It gives a picture of hypertrophy of left upper thigh and upper leg which infact is not a hypertrophy but additional skin fold on left thigh and leg. He has 1 brother, alive and healthy and parents are apparently normal.

On examination of left hip, the greater trochanter is broad and shifted upwards as compared to right side, giving the picture of upwards subluxation of hip. Apparently, all movements of left hip were normal except wide abduction was not possible. There was no tenderness at knee joint but knee joint was hypermobile with marked recurvatum. Patient was walking with hand on knee gait (Fig 3). Left ankle joint was within normal limits. There was no apparent abnormality in spine and rest of his other joints. During ophthalmologic examination multiple Lisch nodules were present on the outer surface of the iris of both eyes. Neurological examination did not reveal any motor weakness or sensory loss in the lower extremities.

All routine blood investigations were within normal limit. Skiagram of pelvis with bilateral hip joints AP view showed degenerative changes in left hip region, marked abnormalities of head of femur. There was proximal migration of head of femur and picture looks as pseudoarthrosis of left hip. Skiagram of left knee (Fig 4) shows marked degenerative changes in the lower end of

femur, upper end of tibia with small patella. Skiagram of spine (Fig 5) showed mild scoliosis and anterior scalloping of vertebrae. Colour and pulsed Doppler study of vessels of left lower limb did not reveal any significant abnormality in the vascular system, however there was redundancy of skin with prominent great saphenous vein and its tributaries. MRI (Fig 6) of left thigh and calf revealed that there is hypertrophy of left thigh and calf with signal intensity alteration and heterogeneity of the muscles and subcutaneous fat of left thigh, and similar intensity alterations in muscles and subcutaneous soft tissues of left inguinal and gluteal region, suggestive of plexiform neurofibromatosis. Left femoral head, neck, greater trochanter and acetabulum are deformed in shape with irregular surfaces and articular surface of left knee joint are deformed with large osteophytes and patellar spur. Nerve conduction study of all peripheral nerves did not reveal any abnormality.

Patient was started on strengthening exercise programme and left side knee brace (Fig 7) was given followed by mobility training. He was than referred to plastic surgery for debulking surgery of left thigh and leg.

Plastic surgeon performed the unusual debulking procedure and removed excessive loose skin around thigh and leg, subcutaneous fascia including extra fat from left thigh and leg. The cutaneous nerves were thickened and vascularity was much more. Proper care was taken



**Fig 1-** Hypertrophy of Left Thigh and Leg **Fig 2-** Multi Cafe-lu Spot **Fig 3-** Hand on Knee Gait during Walking

to preserve delicate cutaneous nerves during debulking procedure and shape of thigh/leg was restored to near normal. The patient was happy and satisfied by outcome of surgical intervention.

Removed specimen was sent for histopathological examination. Three specimen were sent, 1st piece was  $39 \times 9 \times 1$ cm, 2nd piece was  $20 \times 5 \times 11$ cm and 3rd piece was  $27 \times 7 \times 1$ cm. Outer surface showed skin with prominent hair follicles. On microscopic examination, there was proliferation of monomorphic spindle cells, which have fine chromatin with inconspicuous nuclei and abundant cytoplasm with out any atypical cell. The spindle tumour cell infiltrating in adenexal structural and deep within adipose tissue. The above findings are suggestive of diffuse neurofibroma.

### Discussion:

Neurofibromatosis-1 is a hamartomatous disorder, with the genetic defect localised to the long arm of chromosome 17q11.2<sup>7</sup>. The National Institutes of Health (NIH) in 1987 established diagnostic criteria of patients with NF-1. These well-recognised diagnostic criteria are neurofibroma (two or more simple, or one plexiform neurofibroma), cafe-au-lait spots (six or more,  $>5$ mm in greatest diameter in children and  $>15$ mm in adults), Lisch hamartomas in iris (two or more), axillary or inguinal freckling, skeletal abnormalities (sphenoid dysplasias or cortical thinning, with or without pseudoarthrosis), optic glioma and first-degree relative with NF-1. Presence of two or more of these seven criteria establishes the diagnosis of NF-1.<sup>8</sup> In our patient, three of the seven above mentioned diagnostic features were present.

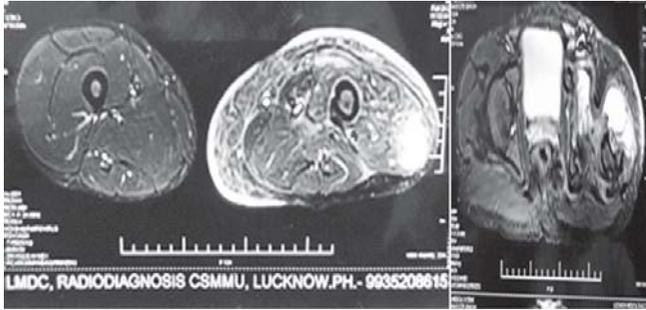
Plexiform neurofibroma, found in up to 26.7% of patients with NF-1 is considered an uncommon skin tumour<sup>5</sup>, usually presenting at birth or during the first several years of life. They are non-encapsulated, poorly circumscribed tumours that diffusely infiltrate the nerve and the adjacent fat and muscle. As a result, neurofibromas are usually unresectable tumours, where tumour resection is impossible without sacrificing the nerve tissue. Fusiform enlargement of multiple nerve fascicles and branches are characteristic. Plexiform neurofibromas contain a mixture of Schwann cells, fibroblasts, reticulin and collagen fibres and a loose mucoid matrix interspersed between the axons of the parent nerve. They typically affect the trunk and extremities, but may also involve the head-neck and bladder. Associated bone dysplasia is often encountered secondary to chronic hyperaemia or



**Fig 4-** Showing Proximal Migration of Femoral Head and Deformation of Acetabulum, Head of Femur and Distal Femur



**Fig 5-** Anterior Scalloping of Vertebrae with Mild Scoliosis



**Fig 6-** MRI Left Thigh-hypertrophy of Left Thigh and Calf with Signal Intensity Alteration and Heterogeneity of the Muscles and Subcutaneous Fat of Left Thigh



**Fig 7-** Patient with Knee Orthosis

as part of the mesodermal dysplasia. Such tumours give rise to a variety of problems, including disfigurement and functional impairment.<sup>9</sup>

### Summary:

We describe a rare case of plexiform neurofibromatosis, presenting as progressive hypertrophy of left thigh and leg, its typical clinical, microscopical, and imaging features diagnostic of NF1. We also propose the proper and meticulous clinical assessment in such cases for early detection of NF1 followed by proper debulking surgical procedure by an experienced plastic surgeon.

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## REHAB CHALLENGES

A thirty year male patient suffered from open fracture of his left femur due to a massive road traffic accident. The wound (Fig 1) was ugly looking due to infection which was treated with intravenous antibiotic and blood transfusion in the department of Orthopaedics. The orthopaedic team was unable to do any open reduction and internal fixation due to the primary wound infection. Subsequently his wound was healed and partial weight bearing was started after development of some callus formation (Fig 2).

When the patient attended PMR OPD his knee and hip ROM were full and he was pain free. He was managing a sensible gait (Fig 3) with bilateral axillary crutches. But he wants to leave his crutches at this juncture (Fig 4).



**Fig 1-** X-ray after Wound Healing



**Fig 2-** Callus Formation



**Fig 3-** Good Standing Balance



**Fig 4-** Last X-ray Showing Callus and Malunion

## REHAB QUIZ

1. **In normal walking the pelvic tilt is about**
  - A) 2 degree
  - B) 5 degree
  - C) 7 degree
  - D) 10 degree
2. **In dislocation of the hip, which of the following nerves is commonly injured?**
  - A) Sciatic
  - B) Femoral
  - C) Superior gluteal
  - D) Obturator
3. **F wave amplitude is**
  - A) Larger than M wave
  - B) Larger than H wave
  - C) Smaller than S wave
  - D) None of the above
4. **Which of the following sensations requires the parietal lobe for final perception?**
  - A) Graphesthesia
  - B) Pain
  - C) Position sense
  - D) Pressure sense
5. **Which of the following conditions has the best prognoses?**
  - A) Amyotrophic lateral sclerosis
  - B) Werdnig- Hoffmann disease
  - C) Myotonica atrophica
  - D) Myotonica congenita
6. **Suction sockets can be best used by**
  - A) Geriatric amputees
  - B) Young traumatic amputees
  - C) Any age group amputated for diabetic gangrene
  - D) Patients with previous bypass surgery with a new amputation.
7. **Rigid postoperative dressing allow the amputee all except to**
  - A) Obtain a permanent prosthesis earlier.
  - B) Use crutches sooner
  - C) Improve balance sooner
  - D) Bear weight earlier than with an immediate prosthesis fitting.
8. **The following laboratory findings are characteristic of Poly myalgia rheumatic except**
  - A) Anemia
  - B) Absent rheumatoid factor
  - C) Rise in CPK level
  - D) Elevated fibrinogen level
9. **Ultrasonic effect is primarily due to**
  - A) Slowing of the rate of diffusion of ions across the biologic membranes
  - B) Elevation of temperature from absorption of energy
  - C) Change in permeability of the membrane potential
  - D) Gaseous cavitation.
10. **All of the following conditions may due to enzyme deficiency, are characterized by inheritance, or are confused with Duchenne's disease except**
  - A) McArdle's
  - B) Pompe's
  - C) Thomsen's
  - D) Central core myopathy

## ANSWERS

September 2013 issue:

1-A; 2-C; 3-C; 4-C; 5-A; 6-C; 7-B; 8-B; 9-A; 10-C

## Urodynamic study of Bladder Behaviour in Traumatic Spinal Cord Injury Patients in Response to Rehabilitation

Singh Ningthoujam Jungindro<sup>1</sup>, Keshkar Sanjay<sup>2</sup>,  
Singh Naorem Ajit<sup>3</sup>, Kumar Ratnesh<sup>4</sup>

### Abstract

**Objective:** To study the bladder behaviour in response to rehabilitation intervention in traumatic spinal cord injury (SCI) patients using urodynamic study (UDS) and to compare the nature of the bladder in a rehabilitated and non-rehabilitated neurogenic bladder of traumatic SCI.

**Study design:** Prospective follow-up study.

**Setting:** Rehabilitation ward of National Institute for the Orthopaedically Handicapped.

**Material and Method:** Thirty traumatic SCI patients mean age  $31.4 \pm 7.9$  years, 26 males and 4 females, admitted for rehabilitation were done UDS to see the bladder behaviour in response to rehabilitation intervention and compare the nature of bladder of the rehabilitated and that of the non-rehabilitated neurogenic bladder. Rehabilitation of the bladder was done at least for three months. The study lasted for 2 years with a minimum of one year follow-up.

**Result:** In upper motor neuron (UMN) neurogenic bladders there were significant changes in the max. cystometric capacity ( $p=0.018$ ) after rehab intervention. The compliance, Pdet. at first desire to void and Pdet. at max. cystometric capacity of these patients were also found to have significant correlations ( $p=0.012$ ,  $0.010$  and  $0.014$  respectively). But the volume at the first desire to void does not show must significant changes after rehab intervention ( $p=0.45$ ). Significant reduction of amplitude and frequency was found in involuntary contractions (detrusor hyper-reflexia). In similar comparison of the lower motor neuron (LMN) neurogenic bladder significant changes after rehab intervention, could be found only in the max. cystometric capacity ( $p=0.018$ ). Other variables like compliance, volume at first desire to void, Pdet. at first desire to void, and Pdet. at max. cystometric capacity were found to have no significant changes after rehab intervention ( $p=0.168$ ,  $0.194$ ,  $0.324$ ,  $0.302$  respectively).

**Conclusion:** The change in the nature of the rehabilitated neurogenic bladder is different with the type of bladder.

**Key words:** Traumatic spinal cord injury, urodynamic study, neurogenic bladder.

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### Introduction:

Spinal cord injury has been known since thousands of years back i.e. since the Hippocrates' time. But its management has improved only few decades back, by focusing primarily on better bladder management. It is difficult to obtain Indian statistics of SCI due to difference in data collection. In one of the studies it is approximated to 20,000 new cases added every year.<sup>1</sup> According to the National Spinal Cord Injury Statistical Centre (NSCISC), approximately 12,000 new cases are added each year in U.S.<sup>2</sup> In developing countries, the road traffic accident and fall from height seem to be the most common cause of spinal fractures and spinal cord injuries.<sup>1</sup> The nature of injury varies with age, where motor vehicle crash is the leading cause of SCI until age 45; however, falls represent the leading cause of SCI in

age group 46 and above. Recreational sports and acts of violence decrease with advancing age as cause of injury. Men suffer traumatic SCI much more commonly than women, at a 4.2:1 ratio<sup>2</sup>.

Renal failure arises as a result of bladder and sphincter dysfunction, infection due to stasis and amyloid disease, are most common cause of mortality in chronic SCI till 1970s<sup>3</sup>. Over the past 40 years long term survival of the spinal cord injured patient has improved due to development of expertise units which has prevented lethal complications of pressure sores, respiratory and urological problems<sup>4</sup>. Unfortunately, despite these advances, permanent disability and problems with bladder still exist. Since urinary tract complications are related to altered bladder and sphincter function it means complications are preventable many a times. The urodynamic investigation has important role to know detrusor and sphincter status of the patient and plan its management accordingly<sup>5,6</sup>. In the present study UDS is used to assess the manifestations of the bladder following SCI. UDS is done in two groups of SCI patients, one who have not done any form of bladder rehabilitation and the other who have undergone a standard bladder rehabilitation programme.

## Materials and Methods:

The study was conducted on thirty inpatients, admitted for institution based rehabilitation (IBR) in the rehabilitation ward of the National Institute for the Orthopaedically Handicapped, Kolkata as part of post-graduate training with due approval of the institute's ethical committee. The study was conducted for a period of 2 years (August 2007 to July 2009), and a minimum follow-up period of one year. The study included only the traumatic causes of SCI. A detailed clinical history, examination and a baseline investigation were taken for all patients. Written consents were taken after proper instruction given about the procedure of UDS. The cases were divided into two groups, (i) those who had never received any specific rehabilitation programme for bladder or were simply left with an indwelling catheter and (ii) those who had undergone rehabilitation programme of the bladder for at least 3 months, according to its type of manifestation in UDS with the available treatment protocols<sup>3,7</sup>. Patients who came to us without any bladder rehabilitation for 3 months or more since the time of injury were included in the first group. Patients who came earlier were included in the latter group. UDS was performed and treated according to the

findings. Treatment includes catheterisation, intermittent clamping, intermittent catheterisation, behavioural therapy and oral medications (oxybutynin or tolterodin) depending on the nature of the bladder. Interventions like botulinum toxin injection of detrusor were not included.

As part of patient preparation they were given two tablets of dulcolax (bisacodyl 5mg) or in suppository form (10mg) on the night before the test. Next morning on the day of examination they were asked to void their bowels and not to take any solid food till the test is over. UDS was done and recorded for both filling and voiding cystometry using multichannel auto pumping urodynamic machine (Medtronic DUET<sup>®</sup> LOGIC G|2, Software: IEC Publication 60601-1-1). Normal saline at room temperature was used for infusion at the rate of 50 ml per minute. All the patients were instructed to take ciplox 500 mg (ciprofloxacin) just after the test and continued for 3 days two times daily.

For comparison basis we have divided the cases into four main groups viz, rehabilitated upper motor neuron, rehabilitated lower motor neuron, non-rehabilitated upper motor neuron and non-rehabilitated lower motor neuron cases. To compare the changes of the quantitative variables after rehabilitation in comparison to the non-rehabilitated, unpaired Student's t-test was used. The result was considered significant at 5% ( $p < 0.05$ ) level of significance. The analysis was carried out using statistical package "SPSS V7.5".

## Results:

Out of the 30 patients, 86.7% (n=26) were males and 13.3% (n=4) were females. The mean age group was  $31.4 \pm 7.9$  (range 17- 45 years). Maximum number of patients belong to the age group 26–40 years (60%, n=18) (Table 1).

Road traffic accident (RTA) was the most common cause (50%, n=15) followed by fall from height (23.3%, n=7). Majority of the patients were daily labour (23.3%, n=7). The mean duration of stay in the rehabilitation ward was  $75.16 \pm 29.4$  days (range between 16 and 145 days).

Nineteen patients (63.33%) have UMN type of neurogenic bladder of which 17 (56.65%) have detrusor hyper-reflexia with detrusor sphincter dyssynergia (DSD) and 2 (6.66%) without DSD. Of the 19 patients, 18 were of suprasacral spinal cord lesion, including cervical (4), thoracic (12), lumbar (2) and 1 sacral segment. Eleven patients (36.66%) have LMN type of neurogenic bladder

of which 4 (13.33%) were of lumbar segment lesion and 7 (23.33%) of sacral segment lesion (Table 2).

Of the 19 UMN type bladders, 9 (30%) were of rehabilitated and 10 (33.33%) were non-rehabilitated bladder. Among the 11 LMN types, 6 (20%) were of rehabilitated and 5 (16.66%) were non-rehabilitated (Table 3). Comparison of the different variables of cystometry among the rehabilitated and non-rehabilitated was done using unpaired t-test. The values are summarised in Table 4. Another important variable that cannot be represented numerically but can be seen graphically is the involuntary contractions of the bladder (detrusor hyper-reflexia).

### Discussion:

Neurogenic bladder (NB) following traumatic SCI is

unavoidable and the management of NB dysfunction is a crucial component of a rehabilitation programme. It is difficult to predict bladder and sphincter behaviour on the basis of clinical somatic neurological deficits<sup>5,6</sup>. The purpose of the urodynamic evaluation of SCI patients is to identify those at risk of developing urological sequelae and to determine the requirement of early intervention. The principal goals of managing NB are to preserve renal function and to maintain patient’s quality of life (QoL) by decreasing urological complications. With a proper bladder management method that optimises both renal function and social functioning, the person with SCI can enjoy a much healthier life. Various approaches to managing urinary disorders have been developed in recent years, ranging from surgery to the ingestion of active drugs and clean intermittent catheterisation (CIC) or the insertion of endourethral prosthetic devices<sup>7,8</sup>.

**Table 1:** Population Distribution

Variables		Rehabilitated (n=15)	Non-rehabilitated (n=15)	Total	
Age (yr)	15 - 25	3 (10%)	4 (13.33%)	07(23.3%)	
	26 – 40	10 (33.33%)	8 (26.66%)	18(60%)	
	41 - 60	2 (6.66%)	3 (10%)	05(16.7%)	
Sex	Male	14 (46.66%)	12 (30%)	26(86.7%)	
	Female	1 (3.33%)	3 (10%)	04(13.3%)	
Cause	RTA	9 (30%)	6 (20%)	15(50%)	
	Fall from height	4 (13.33%)	3 (10%)	07(23.3%)	
	Slipped	2 (6.66%)	2 (6.66%)	04(13.3%)	
	Violence	0	4 (13.33%)	04(13.3%)	
Occupation	Daily labour	4 (13.33%)	3 (10%)	07(23.3%)	
	Student	2 (6.66%)	4 (13.33%)	06(20.0%)	
	Business	2 (6.66%)	3 (10%)	05(16.6%)	
	Farmer	2 (6.66%)	1 (3.33%)	03(10.0%)	
	Unemployed	2 (6.66%)	0	02(6.6%)	
	Housewife	0	2(6.66%)	02(6.6%)	
	Others	3 (10%)	2 (6.66%)	05(16.6%)	

**Table 2:** Level of Injury and Urodynamic Findings

Level of Injury (No. of patients)		UMN type bladder		LMN type bladder areflexia (%)	Normal
		Detrusor hyper-reflexia with DSD (%)	Detrusor hyper-reflexia without DSD (%)		
Suprasacral spinal	Cervical (4)	4 (13.33)	×	×	×
	Thoracic (12)	11 (36.66)	1 (3.33)	×	×
	Lumbar (6)	2 (6.66)	×	4 (13.33)	×
Sacral (8)		×	1 (3.33)	7 (23.33)	×

Though the study was not intended to see the incidence or prevalence of SCI, we have shown here simply to highlight its correlation with the international data. According to the NSCISC, the incidence rates are lowest for the paediatric age group and increase with age with the average age of 40.2 at the time of injury<sup>2</sup>. In our study majority of the cases belonged to age group 26 to 40 (59%) followed by the age group 15 to 25 (22%). The proportion of men in the NSCISC database is 80.8% making a gender ratio of 4.2:1. In our study male patients comprised 86.7% making a gender ratio of 6.5:1.

Natures of cause of SCI as per NSCISC database are many in which the motor vehicle crashes rank first (41.3%), followed by falls (27.3%), violence (primarily gun shoot wounds) (15.0%), sports injury (7.9%), and others (8.5%) including hit by a falling objects, medical or surgical complications, pedestrians being stuck by motor vehicles, stab wounds, bicycle mishaps, and violent personal contacts etc. In our study majority of the cases were due to road traffic accident (50.0%),

followed by fall from height (23.3%), slipped (13.3%) and violence (13.3%).

In our study most of the patients belonged to daily labour comprising 23.3% followed by students (20%) and business persons (16.6%). Farmers constitute about 10.0%. Housewives and unemployed persons constitute 13.2% (6.6% each) and others 7% (include driver, sports person etc.)

There is always controversy of using antibiotics as prophylaxis during urodynamic study. Some studies do not favour of giving antibiotic but most of the studies recommend antibiotic prophylaxis. Quek and Tay<sup>9</sup>, 2004, in their study of 93 patients (44 males and 49 females) found that significant bacteriuria after urodynamic pressure flow study is largely asymptomatic and self-resolving. There was extremely low rate of symptomatic infection and hence antibiotic prophylaxis was not recommended<sup>9</sup>.

Many of the studies recommend use of antibiotic. Pannek and Nehiba<sup>10</sup> 2007, Lathe *et al*<sup>11</sup> 2008, Bergman and McCarthy<sup>12</sup> 1983, Kartal *et al*<sup>13</sup> 2006 are in favour of antibiotic use before or after UDS. In our study we use ciprofloxacin 500mg two times daily for 3 days.

In our study we could see that some of the bladders behave in contrary to prediction from the level of spinal injury. Four (13.33%) of the 22 suprasacral spinal injury show hyporeflexic/areflexic bladder and one (3.33%) of the 8 sacral spinal injury show detrusor hyper-reflexia without DSD. Similar findings were seen by Kaplan *et al*<sup>14</sup> 1999 where 20 of 117 cervical cord lesions had

**Table 3:** Distribution of Neurogenic Bladder to Rehabilitated or Non-rehabilitated

Patients	Types of Bladder		Total
	Upper motor neurone	Lower motor neurone	
Rehabilitated	9	6	15
Non-rehabilitated	10	5	15
Total	19	11	30

**Table 4:** Unpaired Student's Test

Urodynamic Study variables	UMN type bladder			LMN type bladder		
	Rehabilitated (mean)	Non-rehabilitated (mean)	p-value	Rehabilitated (mean)	Non-rehabilitated (mean)	p-value
Max. cystometric capacity	524.55 ± 183.01	340.80 ± 118.90	p=0.018	511.00±79.78	306.00±70.71	p=0.018
Compliance	15.17±10.05	5.39±2.7	p = 0.012	15.92±9.90	3.9±2.5	p=0.168
Pdet. at first desire to void	24.89±23.86	51.89±23.86	p = 0.010	25.4±29.91	51.00±18.38	p=0.324
Pdet. at max. cyst. capacity	40.78±25.95	67.40±15.59	p= 0.014	38.83±30.33	65.00±15.56	p=0.302
Vol. at the first desire to void	294.78±149.45	251.56±80.45	p=0.45	314.2±58.22	247.50±23.33	p=0.194

areflexia, 26 of 84 sacral cord had either detrusor hyper-reflexia or detrusor-external sphincter dyssynergia. Light and Beric<sup>15</sup> 1992 explained the unexpected bladder behaviour in high spinal cord injury to be suspicion of associated lesion or dysfunction of sacral cord. Such poor correlation between physical finding and level of injury is also shown in many other studies<sup>5,6</sup>.

Literature shows limited evidence on similar study that compares the different variables of UDS in a rehabilitated and non-rehabilitated NB. In a prospective study by Khanna *et al*<sup>16</sup> 2009, a total of 82% patients underwent three to four urodynamic studies which revealed an increase in cystometric capacity and a decrease in the maximum detrusor pressures. In our study significant changes occur in the max. cystometric capacity, compliance, Pdet. at first desire to void, and Pdet. at max. cystometric capacity in UMN type of bladder but insignificant correlation in volume at the first desire to void after rehabilitation. During the graphical analysis we also found that there were significant reductions in the involuntary contractions (detrusor hyper-reflexia), not amounting to leakage, which occurs during the filling cystometry. It cannot be quantify in numeric.

In similar comparison of the LMN type bladders, significant correlation could be found only in the max. cystometric capacity. Other variables like compliance, volume at first desire to void, Pdet. at first desire to void, and Pdet. at max. cystometric capacity were found insignificant.

### Conclusion:

The change in the nature of the NB after rehabilitation is different with the different types of bladder and not all variables of the cystometry changes significantly after rehabilitation. The study population in the present study is small and may require further study with a larger population.

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# A Multidisciplinary Rehabilitation Approach for Writer's Cramp: A Case Study

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### Abstract

A 61 year-old male was referred from neurology to PMR department RML hospital, New Delhi, with difficulty in writing for 2 years prior to consultation. Initially difficulty in writing which is accompanied with pain after prolonged writing. Also he noticed tremor and stiffness while writing. Disability progressed and he was unable to write even a few words legibly and cannot hold object (spoon) which leads to anxiety and dependency.

When the patient was examined at PMR OPD, EMG/NCV study was advised, which showed normal sensory and motor nerve functions.

The present treatment involved the use of a multidisciplinary therapeutic approach with Physical Medicine and Rehabilitation Specialist, Clinical psychologist and Occupational Therapist which gave us satisfactory results within 6-8 months. The findings in this case is very encouraging and studies with large sample sizes can be considered for further conclusive evidence on the treatment of writer's cramp.

**Keywords:** Writer's cramp, multidisciplinary approach.

### Introduction:

Writer's cramp is classified as an occupational neurosis and is one of a large group of functional motor disorders. Writer's cramp is characterised by muscular spasm of the fingers and hand of the writing arm, often spreading to the muscles of the lower and upper arm and to the shoulder girdle with consequent inco-ordination and discomfort, variously described as fatigue, weakness, stiffness or pain, when attempting to write. Accompanying tremor and jerking of the limb while writing. The pen is then grasped more and more

tightly and the hand writing becomes progressively more illegible up to a stage when writing may become impossible.

**Epidemiology:** It is more common in males and appears most often in the third and the fourth decade. Those involved in constant writing typing and key board telegraphy have higher incidence. In Australian study: prevalence of 14% for writer's cramp in a group of 516 male telegraphists<sup>1</sup>. In an Indian study Mahendru *et al*<sup>2</sup> (1981) have reported a prevalence of 5.4 per thousand among office workers.

### Case Report:

**Case:** A 51 years old male patient referred from neurology department to PMR department of Dr. RML Hospital who was clinically normal except difficulty in writing for 2 years prior to consultation. Initially he had difficulty in writing which is accompanied with pain after prolonged writing, also he noticed tremor and stiffness while writing. Disability progressed and he was unable to write even a few words legibly and could not hold objects (spoon) which leads to anxiety and dependency. There is no history of similar episode in the past. Family history and drug history is not significant. No H/O of HTN, DM, CVA, AVM Basal ganglia or cortical tumour. EMG/NCV study was advised, which showed normal sensory and motor nerve functions.

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**General physical examination:** Assumption of dystonic posture during writing (semiflex position of finger with hyperextension of wrist) was noted. Tremor was present when hand is outstretched. There is no sensory and motor deficit. Biochemical analysis and MRI is normal.

**Pathophysiology:** At present three general lines of work that may indicate physiological substrate for dystonia. All three are persuasive and it is not clear whether they are related to each other or whether one is more fundamental than the other

**Loss of inhibition-**Hallett, 2004<sup>3</sup> found the principal finding in focal dystonia is that of loss of inhibition. Excessive movement causing long bursts of EMG activity, cocontraction of antagonist muscle and overflow of activity into muscle not intended for task<sup>4</sup>.

**Abnormal plasticity-**Repetitive activity over long period seems to be a trigger for its development (an animal model support this idea).

**Abnormal sensory function-**Clinically not demonstrable but can be demonstrate by somatosensory evoked potential (SEP) testing.

The most acceptable hypothesis for the genesis of writer’s cramp is that it starts as a symptom of anxiety neurosis in people whose main occupation involves the use of fingers.

**Treatment plan** (Figs 1-4): The present paper describes the method of treatment usually followed in our department in the management of writer’s cramp (Figs 5 and 6) combining the pshycological and occupational therapy regimes.

- 1) Jacobson relaxation exercises, 2) Desensitisation, 3) Retraining.

**Treatment Sessions**

- 1, 2, 3, 4
- 5, 6
- 7, 8
- 9, 10
- 11,12
- 13, 14
- 15, 16
- 17, 18
- 19, 20
- 21

**Task**

- Learning of relaxation technique
- Supinator writing in liquid approximate 20 cms circles
- Reduction of circle size in semi solid
- Writing individual letters and then words on single ruled paper
- Switching to pronator (pen between index and middle fingers) writing position
- Writing of words and sentences with bigger diameter pen
- Writing with smaller diameter pen
- Writing on four ruled paper to reduce letters to 1 cm (normal) size.
- Writing on blank paper
- Writing with time limit to improve speed

**Treatment duration**

- Each session for 5days and time duration of each session is 40 minutes.
- Per day 2 sessions carried out one sessions with Occupational therapist and second sessions at home.

**Treatment outcome**

Pain, tremor and spasm is symptom free. Patient writing speed is 5-6 words per minute. Symptom free writing more than 200 words and symptom free period 2 years.

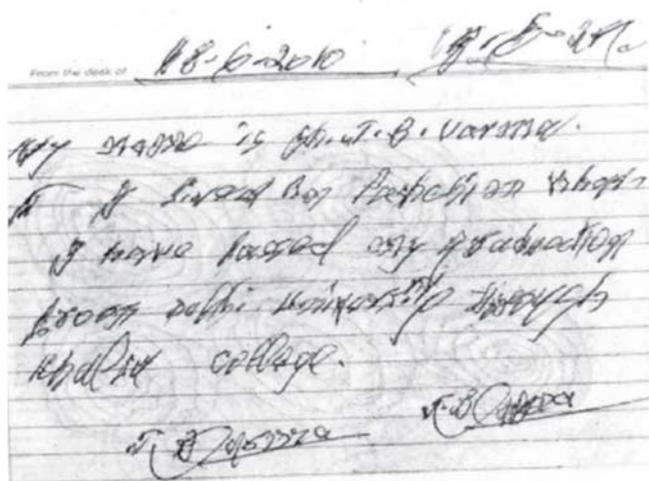


Fig 1- Before Treatment

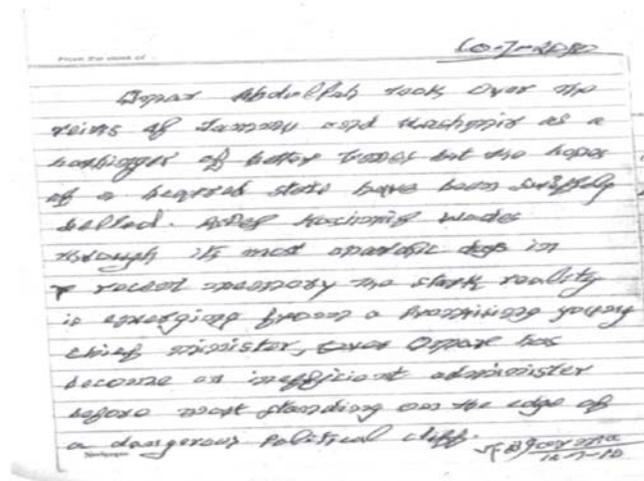


Fig 2- 1st Month of Treatment

27-10-2010

From the desk of

Wizender Sobraj called him the greatest sponsor India has after Sunil Gavaskar and while the jury is out on it, there is no denying that Gauteam Gaumbir made consistency + features of his batting.

Recently though, there have been a few niggles which might have contributed to some technical glitches as well, leading to a slow string of poor scores, which has obviously touched the ground for former in Gaumbir.

The Delhi left-hander is now at the National Cricket Academy in Bangalore not to just prove his fitness but also his technical technique.

T.B. Gaur  
27-10-2010

Fig 3- 3rd Month of Treatment

5-12-2010

From the desk of

An ecstatic Gauteam Gaumbir said after Saturday's game that he never realized he enjoyed leading the side so much. And though he denied the captaincy had anything to do with his current form, he once again displayed comprehensive leadership skills and led from the front with another match-winning century as India notched up an unassailable 3-0 lead in the series against New Zealand - Gaumbir scores only the eighth ODI captain to score back to back centuries as India scored a convincing 9 wicket win in the third ODI at Ranebennur stadium in Vaddesara on Saturday. The end result was a picture of complete contrast with India's total with more than 10 over to spare and Kohli, who remained unbeaten on 63, too was impressive, and finished the proceedings with a huge six over mid-air.

T.B. Gaur  
5-12-2010

Fig 4- 5th Month of Treatment

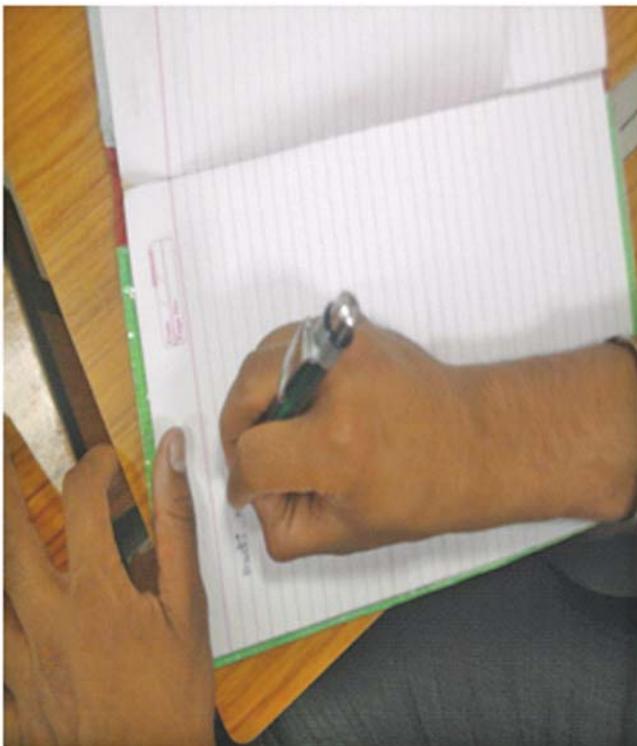


Fig 5: Writer's Cramp

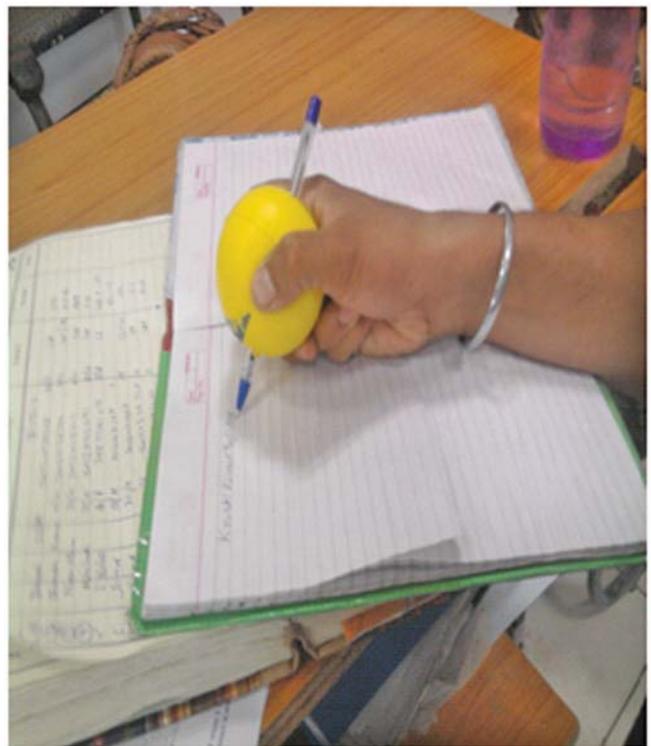


Fig 6: Writer's Cramp

**Discussion:**

The main purpose of this paper is to say that in our treatment plan [the combined approach of i) relaxation exercises ii) desensitization and iii) retraining exercises] usually followed in our department is very useful patients presenting with writer's cramp (traditionally known to

be refractory to several therapeutic approaches) as the duration of treatment is less and no recurrence has been found.. No attempt was made to separate the treatment components. Nor is any attempt made to make a comparative evaluation of this treatment plan.

Several treatment modalities have been tried by various

workers. The view that radicular irritation of cervical spine leads to treatments like galvanic and faradic stimulation, electrified pens, cortisone, ultrasonic therapy and cervical traction. But these treatments were of no significant benefit. Some neurologists, viewing the disturbance as being related to extrapyramidal disorders, have used antiparkinsonism drug, with poor success. Some psychiatrists viewed it as a hysterical symptom while others viewed it as an obsessive neurosis. Sedation, hypnosis and psychodrama were tried unsuccessfully.

A psychosomatic formulation states that the act of writing is a refined and delicate motor skill, incompatible with grosser postures of the upper limb, associated with emotional states like anger. When such a state affects a person chronically or arises specifically in relation to the act of writing, it may progressively distort the writing as the person makes a succession of attempts to overcome the difficulty. Crisp and Moldofsky<sup>5</sup> reported the usefulness of relaxation and re-educative techniques along with psychotherapy in the treatment of writer's cramps. Janet *et al*<sup>6</sup> 1925 was advocating a complex programme of exercises to strengthen the extensor of the hand followed by teaching the patient to write with the hand supinated to encourage activity of the extensor muscles and to discourage excessive flexion of the digits. Then, to redevelop accurate writing movements of the hand, such shaping devices as special keyboards and pigeonholes were introduced.

More recently relaxation has been advocated in the treatment of the condition<sup>7</sup> and in one study<sup>5</sup> it was used in conjunction with a programme in which the individual

was taught to write again, using in the first instance a pen with a wide, soft, felt nib for the drawing of simple shapes and progressing to adult script with a normal pen. While general muscular skeletal relaxation<sup>8</sup> was the aim, particular emphasis in this respect was placed on the upper limbs.

### Conclusion:

It's a fascinating condition. Recent findings have been useful in suggesting new therapeutic approaches to the disorder. Present case study demonstrate that writer cramp can be treated using a multidisciplinary approach. Finding in this case is very encouraging and study with large sample size is being done for further conclusive evidence on the treatment of writer's cramp.

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