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Association of gleno humeral joint subluxation and complex regional pain syndrome in hemiplegic patients

KEYWORDS

Complex Regional Pain Syndrome; Shoulder pain; Shoulder subluxation; Stroke.

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ABSTRACT

Background: Complex regional pain syndrome is a common condition found in patients with cerebro vascular accidents which is associated with significant morbidity. Shoulder joint disruption is one of the reasons attributed to this pathology. The primary objective of this study was to find out any association between glenohumeral joint subluxation and chronic regional pain syndrome in hemiplegic patients. Secondary objective was to find the correlation between the degree of shoulder pain and grade of subluxation in these patients. Methods: This was a Case Control study, conducted from January 2015 to December 2015, at Government Medical College Hospital, Trivandrum. Study population included hemiplegic patients attending the stroke clinic and hemiplegic patients admitted in Department of physical medicine and rehabilitation at Government Medical College. 30 cases and 30 controls were included in the study. Results: Presence of shoulder subluxation was significantly higher in patients with chronic regional pain syndrome. Also presence of pain was significantly higher in the patients with shoulder subluxation than those without subluxation. Significant correlation was found between severity of shoulder pain and grades of subluxation in patients without chronic regional pain syndrome. Conclusions: As per the results of the study, glenohumeral joint subluxation might be one of the factors causing hemiplegic shoulder pain and chronic regional pain syndrome type 1 in hemiplegic patients. Preventive measures would include careful handling of the patient during the acute phase of stroke. Appropriate management of glenohumeral joint subluxation should be instituted in a hemiplegic patient at the earliest to ensure optimal outcome.

Introduction

According to the World Health Organization, stroke or cerebral vascular accident (CVA) represents the second most common cause of death in developed countries.[1] It is also a major cause of disability, as it results in paralysis and cognitive defects in the surviving population. One of the frequent complications of stroke is shoulder pain, with a very high prevalence. It is independent of age and sex and occurs around the second week after the stroke.[2,3]

Complex regional pain syndrome (CRPS) is a relatively recent terminology coined for the syndrome formerly referred to as reflex sympathetic dystrophy, causalgia, Sudeck's atrophy, shoulder hand syndrome, neuroalgodystrophy or reflex neuromuscular dystrophy. It manifests classically as persistent burning pain in a limb, with a region of intense allodynia, hyperalgesia, extreme guarding of the affected limb, reduced range of motion, and objective evidence of local autonomic dysfunction along with trophic changes. Currently CRPS is categorized as CRPS type I when no clear nerve injury is defined, and CRPS type II when associated with clear nerve injury.[4] Stroke is an important cause of CRPS I, and between 30% and 80% of patients following stroke have been reported to develop CRPS I.[5,6,7,8] The high incidence of CRPS I of the upper limbs in patients with stroke and its painful and functional consequences present a problem to specialists in physical medicine and rehabilitation.[9,10]

The patho-physiology of CRPS is complex, and often occurs following a noxious event.[6] During the acute stroke, when the patient is comatose, positioning and passive physiotherapy may result in trauma to the upper limb, including shoulder dislocation or subluxation. Presence of sensory loss, hemi-neglect and visual inattention may result in improper positioning, rendering the limb vulnerable to frequent trauma, which in turn predisposes to development of CRPS I. Occurrence of CRPS I is a major limiting factor of good functional outcome, and if sufficient attention is not given, may result in frozen shoulder, with subsequent deformity of the hand and fingers. The management of CRPSI remains controversial though various pharmacological and non-pharmacological treatments have been tried.[11]

The occurrence and severity of CRPS I are related to the etiology of the stroke and the intensity and the course of the motor deficit, spasticity, and sensory deficits, along with other associated factors. On the basis of these factors, Perrigot et al. designed a prognostic score which gives an estimate of the risk of CRPS I and its severity, and has been found to be valid.[12] This clinical score was established by attributing a value to each of these factors as a function of its strength of correlation with CRPS I.[13,14] However, this prognostic score does not take account glenohumeral subluxation (GHS), unilateral neglect, or depression, which have also been incriminated in CRPS I in the hemiplegic patient.[15,16]

With this background, we attempted to study shoulder joint pathology and CRPS I in patients with stroke. Primary objective was to find out any association between glenohumeral joint subluxation and chronic regional pain syndrome type 1 in patients diagnosed with hemiplegia. Secondary objective was to know if any correlation existed between the degree of shoulder pain and grade of subluxation in non CRPS 1 group.

Methods

The study design was Case-Control study. Study population included hemiplegic patients attending the stroke clinic and hemiplegic patients admitted in Department of physical medicine and rehabilitation at Government Medical College Trivandrum. The study was approved by the Institutional

Volume : 6 | Issue : 12 | December : 2016 | ISSN - 2249-555X | IF : 3.919 | IC Value : 79.96

Review Board and Human Ethical committee of our institution. All subjects had signed an informed consent form(available in local language also) to participate in the study.

Inclusion criteria: Patients with hemiplegia, Age above 30 years.

Exclusion criteria: Patients with bilateral involvement, with multiple attacks, with other shoulder pathology, with brachial plexus injury, with major trauma after stroke, with psychiatric illness, with spasticity greater than grade 2, with history of shoulder injury prior to stroke, with cognitive deficits.

Sample size was set at 30 subjects, calculated from proportion of exposure variable in controls and odds ratio of previous similar study. About 100 consecutive stroke patients admitted in rehabilitation centre and attended stroke clinic in the Department of physical medicine and rehabilitation, Medical college, Trivandrum were evaluated over a period of 12 months. Among these 30 patients were assigned to CRPS group (Cases) and 30 patients to non CRPS group (Controls) according to modified IASP Diagnostic criteria for diagnosis of CRPS, from signs and symptoms obtained from history and clinical examination. Other patients were excluded based on exclusion criteria. Details of study population was collected based on the proforma. It included personal history, history of present episode of stroke, co morbidities, history of symptoms of CRPS according to modified IASP criteria. Then these patients were examined for signs of CRPS Type 1 according to diagnostic criteria. Presence of glenohumeral joint subluxation in the study population was assessed from antero-posterior radiographs of shoulder joint of affected extremity with arm unsupported and centred around glenohumeral joint. Those with shoulder subluxation were assessed for the grade of subluxation using five point categorisation.[17,18]

0: normal, no subluxation(curvature of glenoid fossa parallel to humeral head)

1: v shaped widening(curvature of glenoid fossa opposed by humeral head, but v shaped widening is present)

2: moderate subluxation(inferior subluxation of humeral head, with most superior margin of humeral head not below the line perpendicularly bisecting the line joining the most superior and inferior margin of glenoid fossa)

3: advanced subluxation:(similar to moderate subluxation, but superior margin of humeral head is level with or below the bisecting line)

4: Dislocation, most superior humeral head margin is level with or below the most inferior margin of glenoid fossa.

These data was collected using a self developed proforma. Shoulder pain in the non CRPS group was assessed by visual analogue pain scale of 10 points. Patient was asked to mark in between the end points of the line of 10 cm length. Its left end represented 0,ie, no pain and right end represented most severe pain and marked as 10. And the distance from left end to the point patient marked was measured using scale and classified into mild, moderate, severe pain depending on VAS (Visual analogue scale) score less than 4, 4-8 and more than 8 respectively.

Data collected using proforma and details were entered into Microsoft excel 2013 and analysed with Statistical Package for Social Sciences Software programme (SPSS) ver 18. Qualitative variables were described by percentage. A p value < 0.05 was considered significant. Chi square test was used to compare differences in the percentage of qualitative variables between groups. Spearman's Rank correlation test were applied to test for correlation between variables. The correlation was measured by Spearman's rho. The maximum possible value is 1. Any value closer to one shows positive correlation.

Results

There were 30 cases and 30 controls altogether in our study. Among hemiplegic patients with CRPS 36.6% were below 60 years and 63.3% were above 60 years and in those without CRPS 46.7 % were below 60 years and 53.3% were above 60 years. There was no statistically significant difference in percentage distribution of age between two groups. Among the hemiplegic patients with CRPS 73.3% were males and 26.7% were females whereas in those individuals without CRPS it was 63.3% and 36.7% respectively. Chi square test showed that there is no significant difference between the groups. There was no significant difference in the marital status in percentage distribution between groups.

There was no significant difference between the groups regarding type 2 diabetes status, lipidemic status, hypertension and heart disease. No significant difference in percentage distribution of smokers and alcoholics was found between groups. Between groups there was no significant difference in the side of hemiplegia or regarding the type of stroke, ie, haemorrhagic and ischemic.

Among the hemiplegic patients with CRPS 30% were without any subluxation and remaining 70 % with subluxation [Table 1]. Among the 70%, 16.7% were with grade 1 subluxation, 43.3% with grade 2 subluxation, and 10% with grade 3 subluxation and there was no case of shoulder dislocation among the patients. Whereas in the group without CRPS majority were without subluxation(76.7%), and 13.3% with grade 1 subluxation and 26.7% with grade 2 subluxation and 6.7% with grade 3 subluxation and none with shoulder dislocation. Shoulder subluxation of two groups were compared with Chi Square test and the p value got was less than 0.05(0.003). Shoulder subluxation is most commonly associated with hemiplegic patients with CRPS as the difference is statistically significant.

Among hemiplegic patients with CRPS, shoulder pain was absent for 16.7% and present for 83.3% [Table 2]. Among those with shoulder pain, there was mild, moderate, severe pain in 13.3%, 43.3% and 26.7% respectively. In group without CRPS, majority of patients had no pain ie, almost 70%. p value of this comparison was less than 0.05(0.001). Shoulder pain is significantly associated with hemiplegic patients with CRPS. Correlation between degree of shoulder pain and grade of shoulder subluxation studied using Spearman's rho correlation and the value got was Spearman's rho of 0.331 and p value of 0.074.

In the CRPS group shoulder pain was present in 90.5% of patients with shoulder subluxation and shoulder pain was also present in 66.7 % patients without shoulder subluxation [Table 3,4]. In this comparison p value was not less than 0.05(0.109). Which means there is some positive correlation between shoulder subluxation and shoulder pain in the CRPS group as Spearman's rho is a positive value closer to 1. Degree of shoulder pain and grade of subluxation were statistically compared and the p value got is more than 0.05. So there is no significant difference or relation between degree of shoulder pain and grade of shoulder subluxation in the CRPS group.

In CRPS absent group, when correlation between degree of shoulder pain and grade of shoulder subluxation was assessed by Spearman's rho correlation, the value got was Spearman's rho of 0.555, with a p value of 0.001. Among the non-CRPS group, 5 (71.4%) out of the 7 patients with subluxation and 4 (17.4%) out of the 23 patients without subluxation reported shoulder pain [Table 5,6]. Among the non CRPS group, 71.4 % of patients with shoulder subluxation had shoulder pain and in those without shoulder subluxation, shoulder pain was present in 17.4%. Presence of shoulder pain and shoulder subluxation were compared statistically in the non CRPS group and the p value was 0.006. So, shoulder subluxation is associated with shoulder pain in non CRPS group. Correlation between degree of shoulder pain and grade of subluxation was studied using

Volume : 6 | Issue : 12 | December : 2016 | ISSN - 2249-555X | IF : 3.919 | IC Value : 79.96

Spearman's rho correlation and the value got was 0.55, with a p value of this is less than 0.05(0.001) which indicates positive correlation. This indicates strong association of degree of shoulder pain and grade of subluxation in non CRPS group. In study group of 60 hemiplegic patients among those with shoulder subluxation 85.7% were associated with shoulder pain [Table 7]. And the difference is statistically significant.

Discussion

This was a case control study, where the cases are hemiplegic patients with CRPS and controls are hemiplegic patients without CRPS. Each group consisted of 30 patients. In the study group majority of the patients were above 60 years and were predominantly men. Both cases and controls were comparable with respect to age and gender. Diabetes was present in almost half of the cases and control group, dyslipidemia in almost one fourth cases in both groups, heart disease in one fifth of the patients and hypertension in more than half of the patients in both groups. Other modifiable risk factors like smoking and alcohol were present in about one fourth of the cases and controls. Among hemiplegic patients with CRPS, left side was more affected than right side while in non CRPS group right and left are equally affected. But the difference is statistically insignificant. Infarct is the most common type of stroke on the study group,ie, almost 85-90%. No significant differences were found between the two groups regarding socio-demographic factors.

The presence of shoulder subluxation in the CRPS group was significantly higher than that of the non-CRPS group (p value 0.003). On comparison of cases and controls, statistically significant. association was found between glenohumeral joint subluxation and CRPS. There was no correlation between shoulder subluxation and shoulder pain in the CRPS group. There was no significant correlation existing between degree of shoulder pain and grade of shoulder subluxation in the CRPS group. Presence of shoulder pain and shoulder subluxation were compared statistically in the non CRPS group and the difference was statistically significant. The presence of shoulder pain was significantly higher in the patients with subluxation than in those without subluxation, ie, shoulder subluxation is associated with shoulder pain in non CRPS group. In the non CRPS group, severity of shoulder pain was related to grade of subluxation.

Despite numerous hypotheses, the underlying mechanisms of CRPS are not clearly understood. CRPS can be associated with many precipitating factors like immobilization of the affected limb, trauma to the joint structures, rotator cuff tears, and spasticity of the shoulder musculature. The role played by glenohumeral joint subluxation remains uncertain. The possible mechanisms of subluxation causing CRPS are overstretching of the peri-articular tissues and impingement of the axillary nerve causing shoulder pain.[18,19] Many researchers have reported an association between shoulder pain and subluxation, claiming that antero-inferior subluxation of the humeral head can act as a triggering factor in the hemiplegic patient.[20,21] Some others have reported that no significant correlation exists.[22] Van Langenberghe and Hogan researched on the correlation between degree of pain and grade of subluxation in 48 patients with hemiplegia and found no significant difference in degree of shoulder pain between patients with or without subluxation and no correlation between grade of subluxation and degree of shoulder pain.[17] They concluded that the role of subluxation in producing shoulder pain in hemiplegic patients might be negligible and subluxation must not necessarily be the cause of hemiplegic shoulder pain.

A study by Dursun et al showed that glenohumeral subluxation was more commonly associated with CRPS and the association was proved to be significant statistically.[23] In the non-CRPS group, subluxation and shoulder pain were found to be associated with each other, but without any correlation between the degree of shoulder pain and grade of subluxation. Findings from this study suggest that shoulder subluxation may be a causative factor for CRPS. Similar study conducted by Tepperman PS et al and Chang JJ et al also shows similar results.[24,25] A study by Lo et al and Paci et al also showed significant relation between shoulder subluxation and shoulder pain.[26,27] Suethanapornkul et al concluded that shoulder pain was significantly more frequent in subjects with shoulder subluxation.[28] However, as per Ikai and his associates, results indicated that there was no relation between shoulder subluxation and pain, and adhesive capsulitis is a main cause of shoulder pain. They concluded that correct positioning and shoulder range of motion exercises are advisable in hemiplegic patients with shoulder subluxation.[16] Studies by Wanklyn et al and Roy et al also concluded that shoulder pain was not associated with subluxation.[29,30]

Conclusions

Chronic regional pain syndrome is one of the most difficult conditions to treat. It is the most painful condition among the complications of stroke. It decreases the overall quality of life of many stroke patients. This condition interfere with the patient's rehabilitation Program. It affects general mobility, ambulation training, and self-care activities such as rolling in bed, transfers, upper and lower body dressing, eating, grooming, bathing, and toileting .This condition makes the patient dependent in his activities of daily living.

In the study conducted in our department, the presence of shoulder subluxation was significantly higher in the CRPS group than in the non-CRPS group. In the non-CRPS group, correlation was found between the degree of pain and grade of subluxation, and presence of pain was significantly higher in the patients with shoulder subluxation than those without subluxation. No significant correlation exists between severity of shoulder pain and grades of subluxation in CRPS group while in the non CRPS group shoulder pain severity depends on grades of subluxation.

Our study has some limitations. In the study group selected, functional status of study group was not included. Shoulder subluxation was assessed using a qualitative method and is not much precise. Also, relatively small sample size of the study may have affected the results. Lack of follow up of subjects to document the progression or regression of the condition also is another drawback.

In conclusion, the findings of the study suggests that glenohumeral joint subluxation may be one of the factors causing hemiplegic shoulder pain and CRPS type 1. Preventive measures, including careful handling of the patient should be ensured in a hemiplegic patient as soon as the event is diagnosed. Appropriate management of subluxation and CRPS should be undertaken vigorously with physiotherapy so as to decrease possible complications and improve patients' quality of life.

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Tables

Table1. Distribution of shoulder subluxation among hemiplegic patients with CRPS and without CRPS

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Shoulder subluxation	CRPS type I				Total		χ^2	df	p
	present		absent						
	N	%	N	%	N	%			
No subluxation	9	30.0	23	76.7	32	53.3	13.875	3	.003*
Grade 1 subluxation	5	16.7	3	10.0	8	13.3			
Grade 2 subluxation	13	43.3	3	10.0	16	26.7			
Grade 3 subluxation	3	10.0	1	3.3	4	6.7			
Total	30	100.0	30	100.0	60	100.0			

χ^2 : chi-square value
df: degree of freedom
p : p value
* : p value < 0.05: statistically significant

Table 2. Distribution of shoulder pain among hemiplegic patients with CRPS and without CRPS

Shoulder pain	CRPS type I				Total		χ^2	df	p
	present		absent						
	N	%	N	%	N	%			
Nil	5	16.7	21	70.0	26	43.3	18.989	3	<0.001*
Mild	4	13.3	3	10.0	7	11.7			
Moderate	13	43.3	5	16.7	18	30.0			
Severe	8	26.7	1	3.3	9	15.0			
Total	30	100.0	30	100.0	60	100.0			

χ^2 : chi-square value
df: degree of freedom
p : p value
* : p value < 0.05: statistically significant

Table 3. Distribution of grade of shoulder subluxation and degree of shoulder pain among hemiplegic patients with CRPS type 1

Shoulder subluxation	Shoulder pain								Total	
	Nil		Mild		Moderate		Severe			
	N	%	N	%	N	%	N	%	N	%
No subluxation	3	60	1	25	3	23.1	2	25	9	30
Grade 1 subluxation	0	0	2	50	3	23.1	0	0	5	16.7
Grade 2 subluxation	2	40	1	25	6	46.2	4	50	13	43.3
Grade 3 subluxation	0	0	0	0	1	7.7	2	25	3	10
Total	5	100	4	100	13	100	8	100	30	100

Table 4. Distribution of shoulder subluxation and shoulder pain in CRPS group

Shoulder subluxation	Shoulder pain				Total		χ^2	df	P
	present		absent						
	N	%	N	%	N	%			
Present	19	90.5	2	9.5	21	100.0	2.571	1	.109
Absent	6	66.7	3	33.3	9	100.0			
Total	25	83.3	5	16.7	30	100.0			

χ^2 : chi-square value
df: degree of freedom
p : p value
* : p value < 0.05: statistically significant

Table 5. Distribution of grade of shoulder subluxation and degree of shoulder pain among hemiplegic patients without CRPS type 1

Shoulder subluxation	Shoulder pain								Total	
	Nil		Mild		Moderate		Severe			
	N	%	N	%	N	%	N	%	N	%
No Subluxation	19	90.5	2	66.7	2	40	0	0	23	76.7
Grade 1 Subluxation	1	4.8	1	33.3	1	20	0	0	3	10
Grade 2 Subluxation	1	4.8	0	0	2	40	0	0	3	10
Grade 3 Subluxation	0	0	0	0	0	0	1	100	1	3.3
Total	21	100	3	100	5	100	1	100	30	100

Table 6. Distribution of shoulder subluxation and shoulder pain in non – CRPS group

Shoulder subluxation	Shoulder pain				Total		χ^2	df	p
	present		absent						
	N	%	N	%	N	%			
Present	5	71.4	2	28.6	7	100.0	7.462	1	.006*
Absent	4	17.4	19	82.6	23	100.0			
Total	9	30.0	21	70.0	30	100.0			

χ^2 : chi-square value
df: degree of freedom
p : p value
* : p value < 0.05: statistically significant

Table 7. Distribution of shoulder pain and shoulder subluxation in study group of hemiplegic patients.

Shoulder subluxation	Shoulder pain				Total		χ^2	df	p
	Present		Absent						
	N	%	N	%	N	%			
Present	24	85.7	4	14.3	28	100.0	18.040	1	<0.001*
Absent	10	31.3	22	68.8	32	100.0			
Total	34	56.7	26	43.3	60	100.0			

χ^2 : chi-square value
df: degree of freedom
p : p value
* : p value < 0.05: statistically significant

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Volume : 6 | Issue : 12 | December : 2016 | ISSN - 2249-555X | IF : 3.919 | IC Value : 79.96

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