Introduction
According to the World Health Organization, stroke or cerebral vascular accident (CVA) represents the second most common cause of death in developed countries. 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.1

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.2 Stroke is an important cause of CRPS I, and between 30% and 80% of patients following stroke have been reported to develop CRPS I.3,22 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.4

The patho-physiology of CRPS is complex, and often occurs following a noxious event.5 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 CRPS I remains controversial though various pharmacological and non-pharmacological treatments have been tried.6,7

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.8,9 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.10,11 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.12,13

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 I 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 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 grade 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 I 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.

ABSTRACT
Background: Complex regional pain syndrome is a common condition found in patients with cerebrovascular 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 grade 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 I 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.

KEYWORDS
- Complex Regional Pain Syndrome
- Shoulder pain
- Shoulder subluxation
- Stroke

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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, lipemic 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 there was no shoulder dislocation in this group. 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(0.00)1. 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 obtained 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...
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. The presence of antero-inferior subluxation of the humeral head can act as a precipitating factor for CRPS. 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 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.

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.

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 limb 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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Conflict of interest: None declared.
Ethical approval: Obtained.

Tables
Table 1. Distribution of shoulder subluxation among hemiplegic patients with CRPS and without CRPS
Table 2. Distribution of shoulder pain among hemiplegic patients with CRPS and without CRPS

<table>
<thead>
<tr>
<th>Blood Flow</th>
<th>CRPS type I</th>
<th>No subluxation</th>
<th>Grade 1 subluxation</th>
<th>Grade 2 subluxation</th>
<th>Grade 3 subluxation</th>
<th>Total</th>
<th>χ²</th>
<th>df</th>
<th>p</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>N % N %</td>
<td>N % N %</td>
<td>N % N %</td>
<td>N % N %</td>
<td>N % N %</td>
<td></td>
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<tr>
<td>Present</td>
<td>9 30.0 23 76.7</td>
<td>32 53.3 13.8</td>
<td>75</td>
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<tr>
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Table 3. Distribution of grade of shoulder subluxation and degree of shoulder pain among hemiplegic patients with CRPS type 1

<table>
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<th>Blood Flow</th>
<th>Shoulder pain</th>
<th>No subluxation</th>
<th>Grade 1 subluxation</th>
<th>Grade 2 subluxation</th>
<th>Grade 3 subluxation</th>
<th>Total</th>
<th>χ²</th>
<th>df</th>
<th>p</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>N % N %</td>
<td>N % N %</td>
<td>N % N %</td>
<td>N % N %</td>
<td>N % N %</td>
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<tr>
<td>Present</td>
<td>5 16.7 21 70.0</td>
<td>26 43.3 18.9</td>
<td>89</td>
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</tbody>
</table>

χ²: chi-square value
df: degree of freedom
p: p value
*: p value < 0.05: statistically significant

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

<table>
<thead>
<tr>
<th>Blood Flow</th>
<th>Shoulder pain</th>
<th>N % N % N % N %</th>
<th>P</th>
</tr>
</thead>
<tbody>
<tr>
<td>Present</td>
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<td>21 100.0 2.571</td>
<td>1</td>
</tr>
<tr>
<td>Absent</td>
<td>6 66.7 3 33.3</td>
<td>9 100.0</td>
<td></td>
</tr>
<tr>
<td>Total</td>
<td>25 83.3 5 16.7</td>
<td>30 100.0</td>
<td></td>
</tr>
</tbody>
</table>

χ²: chi-square value
df: degree of freedom
p: p value
*: p value < 0.05: statistically significant

References