

## Reflex sympathetic dystrophy: a retrospective epidemiological study of 168 patients

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**Abstract** This is a retrospective epidemiological study. The objective is to determine the epidemiological characteristics including the patient demographics, etiological factors, duration of symptoms, treatment modalities applied and clinical outcome of the treatment in reflex sympathetic dystrophy (RSD). Medical records of the 168 patients managed in two tertiary hospitals with the diagnosis of RSD that was made according to both IASP criteria and three-phase bone scan were reviewed. The upper limb was affected 1.5 times as commonly as the lower limb. Of the 168 cases, 10.7% were non-traumatic. In 89.3% of the patients, RSD developed after a traumatic inciting event with a predominance of fracture. In 75.6% of the patients, RSD developed due to job-related injuries. The percentage of successful clinical outcome was 72%. The percentage of the patients that did not respond to therapy was 28%. The management period is long and this causes higher therapeutic costs in addition to loss of productive effort. However, response to therapy is good. On the other hand, in approximately one third of the patients, RSD does not improve despite all therapeutic interventions. In addition to compensation costs, this potentially debilitating feature causes RSD to appear as a socioeconomic problem.

**Keywords** Complex regional pain syndrome · Epidemiology · Reflex sympathetic dystrophy

### Introduction

Reflex sympathetic dystrophy (RSD) is one of the chronic pain disorders. Diffuse and non-dermatomal pain is associated with allodynia, vasomotor and sudomotor changes in RSD [1]. It causes loss of productive effort with longstanding management period and unpredictable functional impairments and handicaps in chronic stage. In this regard, RSD might be considered as a socioeconomic problem.

However, in the literature, little data has been published assessing the epidemiology of RSD. With this point of view, we aimed to put forward the epidemiological characteristics of RSD in the patient population admitted to the physical medicine and rehabilitation departments of our two tertiary hospitals.

### Materials and methods

Included in the study, were 168 patients that were evaluated and diagnosed with RSD according to the modified International Association for the Study of Pain (IASP) criteria [2] and three-phase bone scan in two tertiary military hospitals between 2003 and 2006. Data were collected from medical records of the patients retrospectively. Their demographics, medical histories containing etiological factors, prior medical evaluations, therapeutic modalities used, duration of disease, frequency and duration of hospitalization, home rest periods, and final decision on their disease were reviewed.

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Statistical analysis was performed using the SPSS 11.0 Windows package program. Standard descriptive statistics (frequencies, percentages) were used.

## Results

All of the patients were male. The mean age of the patients was  $23.4 \pm 8.4$  years. Affected region was the hand in 67.9% ( $n=114$ ) of the patients and the foot in 32.1% ( $n=54$ ) of the patients. Affected side was the right side in 53.6% ( $n=90$ ) and the left side in 46.4% ( $n=78$ ). Mean duration of symptoms was  $7.6 \pm 9.1$  months. Number of patients that the disease had occurred on the job was 75.6% ( $n=127$ ) and before duty was 24.4% ( $n=41$ ).

Etiologic factors are as follows: 10.7% ( $n=18$ ) were non-traumatic or injury could not be found from the records, 89.3% ( $n=150$ ) or the remaining patients were post-traumatic. Among these: 55.3% ( $n=83$ ) were fracture (operated, non-operated only casting applied), 16.7% ( $n=25$ ) were incisive injury and 28% ( $n=42$ ) were soft tissue trauma (sprain, fall on the limb, etc) (Table 1).

Hospitalization in our departments: once 39.2% ( $n=66$ ), twice 32.1% ( $n=54$ ), three times 26.7% ( $n=45$ ), and four times 1.7% ( $n=3$ ). Treatments applied: physical therapy (including contrast bath, TENS, diadynamia, therapeutic ultrasound, as various combinations) 100%, therapeutic exercises 100%, calcitonine 38.1% ( $n=64$ ), stellar ganglion blocks 23.2% ( $n=39$ ), intravenous regional blocks (Bier) 10.1% ( $n=17$ ), corticosteroids 27.9% ( $n=47$ ), antidepressants (tricyclic antidepressants, serotonin reuptake inhib-

itors) 10.7% ( $n=18$ ), and anticonvulsants (gabapentin) 16.6% ( $n=28$ ) (Table 2).

Mean duration of total hospitalization  $28.6 \pm 17.3$  days. Mean duration of total home rest with home program was  $2.8 \pm 2.5$  months. Final decisions: number of patients who returned to duty was 72.02% ( $n=121$ ), number of patients who accepted as officially disabled because of health problem 27.9% ( $n=47$ ) (Table 1).

## Discussion

RSD is still one of the most important health problems. Remaining as a hard-to-treat disease, it needs longer medical care, so it causes not only higher therapeutic costs but also higher work time loss [3–5]. Moreover, in some patients, RSD could potentially lead to permanent disability and compensations could be paid to some of these patients in whom RSD occurred due to job-related trauma. Because most of these patients are in their economically productive periods of life, this will cause loss of productive effort too. On the other hand, chronic pain and the potential permanent functional loss, regardless of being mild, moderate or severe, could negatively affect a patient's daily living, work life and social relations with other people. Blake et al. reported that 75% of the caregivers of RSD patients had psychological strain and 53% had poor mood [6]. Geertzen et al. reported 17 of 65 RSD patients had changed their occupation and 30% of the RSD patients had had to stop work for more than 1 year [7]. Hence, RSD might be considered as not only an economical but also a social problem for the patients, their families and the community.

Deficiency of sufficient data about the epidemiological characteristics of this disease has led us to construct such a study. This is a retrospective epidemiological study with 168 patients who suffered from RSD and managed in our two tertiary physical medicine and rehabilitation departments. Although the bone scan is not gold standard as an

**Table 1** Various characteristics of patients

	Number of patients	Percentage of patients
Extremity		
Upper (hand)	114	67.9
Lower (foot)	54	32.1
Side		
Left	78	46.4
Right	90	53.6
Inciting traumatic event		
Present	150	89.3
Fracture	83	55.3
Incisive injury	25	16.7
Soft tissue trauma	42	28
Absent	18	10.7
Occurrence		
Before duty	41	24.4
On-the-job	127	75.6
Decision		
Return to work	121	72.02
Officially disabled	47	27.9

**Table 2** Types of treatments and the number of patients that received each therapeutic modality

Type of treatments	Number of patients	Percentage
Physical therapy	100	100
Therapeutic exercises	100	100
Calcitonine	64	38.1
Stellar ganglion blocks	39	23.2
Intravenous regional blocks (Bier)	17	10.1
Corticosteroids	47	27.9
Antidepressants	18	10.7
Anticonvulsants	28	16.6

aid for the diagnosis of RSD, we took into account only the patients whose diagnoses were also confirmed with bone scan and excluded the remaining patients diagnosed with RSD with the aim of being sure of the diagnoses because this was a retrospective study and we could not re-evaluate the patients. The evidences reported by Bruehl et al., which argues against the traditional staging of the disease [8], suggest that duration-dependent staging needs to be justified, so we did not categorized the patients into subgroups with traditional staging.

Mean age for our population is 23.4 years. This is lower than previous studies in which mean ages were reported to be 38 and 47 years with a predominance of females with a ratio of 2.3–4/1 [9, 10]. Almost all of our patients were recruits and all of them were male. There are contradictory observations regarding the predominance of affected extremity and side in literature. We found that the upper limb was approximately 1.5 times as commonly affected as the lower limbs with slightly higher involvement of the right side.

Veldman et al. found no specific identifiable cause in 10% of the RSD cases [11]. Allen et al. reported 6% of RSD patients developed spontaneously and in 6% the inciting event could not be determined. In our population, we found that a total of 10.7% of the patients developed RSD without a trauma or injury could not be found from the records. Most of the RSD develops after a trauma. Previously reported percentages of post-traumatic RSD were ranging between 88% and 100% [9, 10]. We found that in 89.3% of our patients, RSD developed after a traumatic inciting event. Fracture was the most common cause of RSD in our population (55.3%). In addition to supporting them, this percentage was slightly higher than the previous observations. We think that this was because of the nature of our patient population; being recruits they were exposed to trauma more than most of other people. We found that in 75.6% of the patients, RSD developed due to job-related injuries.

The patients received various treatment modalities (physical therapy, therapeutic exercises, intravenous regional blocks, stellar ganglion blocks, corticosteroid, calcitonin, antidepressants, anticonvulsants). Because most of the patients were administered with more than one type of therapeutic modalities in various combinations, we could not evaluate the effects of each modality to the outcome.

There exist some studies regarding the assessment of the cost-effectiveness of the various RSD management modalities in literature [3–5]. The mean hospitalization duration was  $28.6 \pm 17.3$  days and mean duration of home rest with home program was  $2.8 \pm 2.5$  months in our patient group. These periods were corresponding to the period that the patients were out of duty. Other authors reported that for the management of RSD, the cost of physical therapy alone at

12 months interval is €5,741/patient, US \$6,000/patient [3, 12]. We could not determine the exact total cost, however, all of our patients included physical therapy and additional interventions (Table 2) in addition to hospitalization costs, cost of being out of work and compensation costs.

The mean duration of symptoms was  $7.6 \pm 9.1$  months. In our opinion, this is not only because of longer medical care period. There appears a delay for beginning the therapy as a contributing factor. This delay could be caused partly by the nature of our departments (tertiary clinics). Most of our patients were the patients that could not be treated in first-degree or second-degree hospitals and referred to our clinics. Besides, some other probable causes exist. We think that some diagnostic difficulties or referring the patient to irrelevant clinics might play role to some extent in this delay. Although detailed data could not be obtained for all of the patients from the records, we found out that some patients were referred firstly to internists, neurologists and plastic or cardiovascular surgeons with possible diagnoses of Raynaud syndrome, nerve lesion or vascular pathologies. In most of these misdiagnosed patients, RSD had been caused by minor soft tissue trauma or had developed without an apparent trauma.

As long as RSD remains as an unknown mystery with respect to its exact etiology, mechanism, diagnostic aids and remedy, it will continue to be considered as a way of gaining some secondary benefits by some patients. In our opinion, secondary gains must be taken into account by physicians throughout the whole management period of RSD. The thought of secondary gain can be as simple as making their stay in the hospital longer or as serious as gaining unlawful worker compensation benefits. Some patients try to represent themselves as if they have an incurable disease and abuse their disease to be excluded legally from the army, which is an obligatory duty in our country. This is a way of not only exemption from tight and strict duty but also a way of gaining compensation benefit. It is sometimes hard to decide in some resistive cases whether the clinical picture really does not improve despite all therapeutic approaches or some simulations take place. The physician could feel seriously doubtful and fall into a dilemma. We have had experiences of some patients tightly tying their extremities with rope or string, piece of cloth or rubber to increase their edema and make their clinical appearance more serious [13]. This is another aspect of this disease and in our opinion the clinicians should keep this probability in mind.

Some ways of preventing the secondary gain should be considered. Psychiatric approaches might be considered to be integrated more effectively to the multi-disciplinary therapeutic team. Regular psychiatrist or psychologist interviews might be planned throughout the management period in a required frequency. This might help not only

to reduce the secondary gain attempts but also will be helpful for the management of the disease. There are some reports related with the development of RSD on a background of some psychological dysfunctions [14, 15]. Taskaynatan et al. experienced co-existing factitious disorders in some patients with clinically and radiologically diagnosed with RSD [13]. Bruehl et al. reported that the psychological component of treatment could work synergistically with medical and physical therapies to improve function and increase patients' ability to manage the condition successfully [15].

According to the legal rules of our military service, only the patients that suffered from any job-related disease and decided to be unable to continue his duty with his health status have the right to gain compensation. This needs a legal process that proceeds independently from hospitals after the decision. Thus, we could not give the exact number of patients that were compensated.

Although we could not reach any previously reported supportive observation in literature, the general expectation could be that RSD was a chronic, refractory and poorly prognosed disease. In contrast, Zyluk found that 26 out of 30 patients had resolved at 1-year follow-up [16]. Sandroni et al. found that 74% of RSD resolved, reported that most of the RSD were transient and only a minority became severely disabled, and spontaneous resolution was possibly a rule for the majority of RSD patients [10].

In our population, 72% of the patients were decided to return to work. The decision of returning the patient to work does not mean that all of these patients were exactly cured and all of the signs and symptoms of RSD disappeared. Our official decision is related mainly with the patients' clinical status regarding their disease and ability to continue his military duty. Although some patients were absolutely improved, some of them had mild or moderate symptoms that do not prevent them to work on their duties. We could not obtain from the records the exact data of how many patients had fully improved. However, we think that symptom relief at a satisfactory amount as much as not to prevent them from working could be considered as good response to the treatment. In this regard, 72% of the patients might be accepted as clinically successful outcome, supporting these observations. Furthermore, among the types of injury, Sandroni et al. found that fracture was associated with the greatest resolution rate (91%) [10]. More than half of our patients had post-fracture RSD. This could have contributed to our outcome.

On the other hand, we obtained data about the number of patients that we decided as being "officially disabled" (unable to go on working in military service) because of RSD. This decision means that the signs and symptoms of RSD have not improved at a satisfactory degree despite all therapeutic interventions. The percentage of the patients

that were decided to be officially disabled in the interval of this review was 28%. Besides, we could not know the course of the diseases of the patient, which were decided to be officially disabled because their military duties were ended and they continued their medical care in other hospitals. Thus, we could not reach the long-term clinical results of this group and could not know if any of these patients improved after one or more years.

The retrospective design of this study is a limitation. Data were not collected in a standardized way prospectively. To minimize the potential diagnostic mistakes, three-phase bone scan results were also used in addition to the IASP criteria. Another limitation is that the patient population is composed of those that were referred to the tertiary clinic, mostly young recruits. This could misrepresent the whole patient population who experienced RSD at any age and managed in clinics at any degrees of hospitals.

Within the limits of our study, we conclude that RSD is still a major health problem. The most common cause is trauma with predominance of fracture. Hence, for populations or work fields that contain high risk for job-related accidents, some additional preventive precautions should be considered and advised. These can be educational approaches for the personnel or workers and environmental preventive reorganizations around workplaces. Secondary gain is another problem for the clinician in the management of RSD. We suggest that performing psychiatric approaches more effectively could help to reduce this. The management period is long and this causes higher therapeutic costs in addition to loss of productive effort. However, long-term response to therapy is good. On the other hand, in approximately one third of the patients, RSD does not improve despite all therapeutic interventions. In addition to compensation costs, this causes RSD to appear as a socioeconomic problem. The need for searching for more effective ways of treatment is still alive because of the potentially debilitating feature of the disease.

## References

1. Tan AK, Duman I, Taskaynatan MA, Hazneci B, Kalyon TA (2006) The effect of gabapentin in earlier stage of reflex sympathetic dystrophy. *Clin Rheumatol* (in press)
2. Bruehl S, Harden RN, Galer BS, Saltz S, Bertram M, Backonja M, Gayles R, Rudin N, Bhugra MK, Stanton-Hicks M (1999) External validation of IASP diagnostic criteria for complex regional pain syndrome and proposed research diagnostic criteria. *International Association for the Study of Pain. Pain* 81(1–2): 147–154
3. Taylor RS, Van Buyten JR, Buscher E (2006) Spinal cord stimulation for complex regional pain syndrome: a systematic review of the clinical and cost-effectiveness literature and assessment of prognostic factors. *Eur J Pain* 10:91–101
4. Oerlemans HM, Oostendorp RA, de Boo T, van der Laan L, Severens JL, Goris JA (2000) Adjuvant physical therapy versus

- occupational therapy in patients with reflex sympathetic dystrophy/complex regional pain syndrome type I. *Arch Phys Med Rehabil* 81(1):49–56
5. van Dielen HE, Perez RS, van Tulder MW, de Lange JJ, Zuurmond WW, Ader HJ, Vondeling H, Boers M (2003) Cost effectiveness and cost utility of acetylcysteine versus dimethyl sulfoxide for reflex sympathetic dystrophy. *Pharmacoeconomics* 21(2):139–148
  6. Blake H (2000) Strain and psychological distress among informal supporters of reflex sympathetic dystrophy patients. *Disabil Rehabil* 22(18):827–832
  7. Geerzten JH, Dijkstra PU, Groothoff JW, ten Duis HJ, Eisma WH (1998) Reflex sympathetic dystrophy of the upper extremity—a 5.5 year follow-up. Part II. Social life events, general health and changes in occupation. *Acta Orthop Scand Suppl* 279:19–23
  8. Bruehl S, Harden RN, Galer BS, Saltz S, Backonja M, Stanton-Hicks M (2002) Complex regional pain syndrome: are there distinct subtypes and sequential stages of the syndrome? *Pain* 95:119–124
  9. Allen G, Galer BS, Schwartz L (1999) Epidemiology of complex regional pain syndrome: a retrospective chart review of 134 patients. *Pain* 80:539–544
  10. Sandroni P, Benrud-Larson LM, McClelland RL, Low PA (2003) Complex regional pain syndrome type 1: incidence and prevalence in Olmsted County, a population-based study. *Pain* 103:199–207
  11. Veldman PH, Reynen HM, Arntz IE, Goris RJ (1993) Signs and symptoms of reflex sympathetic dystrophy: prospective study of 829 patients. *Lancet* 342:1012–1016
  12. Kemler MA, Furnee CA (2002) Economic evaluation of spinal cord stimulation for chronic reflex sympathetic dystrophy. *Neurology* 59:1203–1209
  13. Taskaynatan MA, Balaban B, Karlidere T, Ozgöl A, Tan AK, Kalyon TA (2005) Factitious disorders encountered in patients with the diagnosis of reflex sympathetic dystrophy. *Clin Rheumatol* 24(5):521–526
  14. Monti DA, Herring CL, Schwartzman RT, Marchese M (1998) Personality assessment of patients with complex regional pain syndrome type I. *Clin J Pain* 14(4):295–302
  15. Bruehl S, Chung OY (2006) Psychological and behavioral aspects of complex regional pain syndrome management. *Clin J Pain* 22(5):430–437
  16. Zyluk A (1998) The natural history of post-traumatic reflex sympathetic dystrophy. *J Hand Surg* 23:20–23