Predisposing Psychological Factors in the Development of Reflex Sympathetic Dystrophy: A Review of the Empirical Evidence

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Review Article

Predisposing Psychological Factors in the Development of Reflex Sympathetic Dystrophy

A Review of the Empirical Evidence

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Abstract:
Objective: To examine the literature for evidence that psychological factors predispose certain individuals to development of reflex sympathetic dystrophy (RSD).

Data Sources: English-language journal articles that described psychological data on patients diagnosed with RSD, identified through Medline search and bibliography reviews.

Study Selection: All studies reporting data on psychological factors in adult, adolescent, or child patients with RSD were included.

Data Extraction: We extracted data regarding psychological factors in patients with RSD and assessed validity of these studies through evaluation against seven basic research criteria.

Data Synthesis: Of the 20 articles reviewed, 15 reported the presence of depression, anxiety, and/or life stress in patients with RSD. However, the methodological quality of these studies was generally poor, with most meeting three or less of the seven validity criteria. In particular, the absence of prospective designs restricts conclusions concerning whether psychological factors are etiologically related to RSD.

Conclusions: The data reviewed are consistent with a theoretical model in which depression, anxiety, or life stressors may influence development of RSD through their effects on α-adrenergic activity. However, conclusions regarding etiological significance of these factors are not possible due to the dearth of high-quality studies. Suggestions for prospective research are described.

Key Words: Reflex sympathetic dystrophy—Anxiety—Depression—Stress—Psychological factors—Etiology.

Reflex sympathetic dystrophy (RSD) is a descriptive term applied to a range of disorders involving persistent pain that is responsive to treatment with sympathetic nervous system (SNS) blockade (1−4). Among the disorders incorporated under this general term are minor causalgia, posttraumatic pain syndrome, Sudeck’s atrophy, shoulder–hand syndrome, reflex dystrophy, reflex neurovascular dystrophy, and algoneurodystrophy (1,5). Given the variety of disorders incorporated under the term RSD, it is not surprising that there is a lack of consensus regarding the proper diagnostic criteria for
identifying RSD (4,6). Despite this lack of diagnostic clarity, RSD researchers generally agree that these syndromes share the feature of SNS involvement in the etiology and/or maintenance of the syndrome (4). One newer concept is sympathetically maintained pain (SMP), a more general term encompassing RSD as well as causalgia and other disorders that exhibit pain related to SNS activity (7). The studies reviewed in this article were generally conducted before the introduction of newer concepts such as SMP, and reflect a variety of diagnostic criteria for RSD. Because determination of the appropriateness of diagnostic criteria for RSD is beyond the scope of this article, this review is limited to those studies in which the authors have used the diagnostic label of RSD.

Although there are no universally agreed-upon diagnostic criteria for RSD, there are a number of commonly reported features of the disorder. The acute stage of RSD (stage I) is characterized clinically by constant burning or aching pain, hypersensitivity to physical stimulation, muscle spasm, and increased hair and nail growth. Vasomotor instability is typical of the acute stage, with periods of unusually warm skin with erythema alternating with periods of cool, clammy, cyanotic skin. In the dystrophic stage (stage II), the skin becomes predominately cool, cyanotic, and sweaty. Additionally, hair loss occurs and the fingernails become brittle and cracked. In the atrophic stage (stage III), the skin becomes thin and shiny, and severe osteoporosis may occur, although the severity of the pain may diminish (1,5). RSD pain often will appear in unusual patterns (e.g., glove or stocking patterns) that are inconsistent with known distributions of nerve fibers (8).

RSD is believed to result from physical trauma, either accidental or surgical, or various disease states (e.g., myocardial infarction), even though the symptoms of RSD may not appear for some time after the suspected trauma (1). The disparity between the minimal apparent trauma and the severe pain experienced by RSD patients often causes physicians to conclude that the patient is psychologically disturbed and emotionally unstable (9). Another difficulty faced by the physician is explaining why individuals with little or no identifiable trauma develop RSD, whereas others with more significant trauma do not develop the disorder. Thus, the question of whether there are specific psychological factors that make an individual susceptible to the development of RSD has appeared repeatedly in the literature (1,8,10).

Disagreement over the role of psychological factors in the etiology of RSD has been evident since the earliest clinical studies appeared. For example, DeTakats (10) concluded that certain personality characteristics were a result of RSD rather than a cause. Others (11) believed that development of RSD resulted from a “constitutional predisposition.” The disagreements between early authors reflect a key issue in understanding the role of psychological factors in RSD: namely, that the personality characteristics that might predispose to development of RSD must be distinguished from the psychological sequelae (e.g., depression or anxiety) of having a chronically painful condition (12).

Numerous investigators have concluded that although there are psychological factors that predispose one to develop RSD, much of the psychological dysfunction associated with RSD is a result of the disorder. For example, Bonica (1) stated that although some patients may be psychologically predisposed to develop RSD, the psychological disturbances in 90% of RSD patients are the result of prolonged pain and disability. Similarly, Poplawski et al. (8) noted that although individuals with psychiatric illness are more prone to the development of RSD, many psychologically stable patients develop emotional instability after prolonged exposure to RSD. Potentially, the identification of specific psychological risk factors for RSD would support more regular use of adjunctive psychotherapeutic care in the treatment of RSD patients and might allow targeting of preventive interventions for these high-risk individuals after sustaining physical trauma. Conversely, empirical evidence that psychological factors are not involved in the development of RSD would suggest that assumptions that all RSD patients are psychologically impaired may need to be modified.

In a recent book chapter, Haddox (13) reviewed the literature regarding RSD and causalgia and concluded that there is insufficient evidence that an “RSD personality” exists. However, Haddox’s review was limited to adults. The primary purpose of this article is to build on Haddox’s review by examining (within a unified theoretical framework) the literature regarding psychological factors in the etiology of RSD in both adults and children. This review will also be specifically focused on the methodological strengths and weaknesses of this literature. The theoretical model underlying this review...
REFLEX SYMPATHETIC DYSTROPHY

will be presented first. The literature regarding psychological factors in RSD in adults will then be reviewed, followed by a review of the literature regarding RSD in children. Because of similar weaknesses among many of the studies reviewed, general criticisms of the RSD literature in adults and children will be offered after the reviews.

THEORETICAL MODEL

Current theorizing regarding the physiological basis of RSD indicates a role for the SNS in the development of the hyperalgesia and hyperpathia components of RSD (14). Some investigators have focused on the afferent activity in mechanoreceptors that can be induced by SNS activity, proposing that this afferent activity evokes sufficient activity in sensitized, spinal, wide dynamic range neurons to produce intense pain (7). Others (3) have proposed a vicious cycle in which nerve lesions result in spinal cord synaptic changes that create abnormal SNS outflow. Afferent terminals in the area of the lesion are then believed to become hypersensitive to this abnormal SNS activity, resulting in the sensation of pain that is sympathetically maintained. In sum, RSD is believed to involve both peripheral and central mechanisms (15).

The theories described above and other related research indicate that the RSD syndrome, particularly the hyperalgesic component, is related to α-adrenergic activity (4). It is interesting to note that disturbances in α-adrenergic activity have been described in the clinical syndromes of anxiety and depression. For example, the generalized anxiety symptoms common to both panic disorder and depression have been shown to be associated with decreased α-adrenergic receptor responsiveness to clonidine (16). Life stress in the preceding 6 months has also been related to decreased α-adrenergic receptor responsiveness (16). Decreased receptor responsiveness is consistent with receptor downregulation resulting from chronic α-adrenergic hyperactivity. We theorize that for a person with a nerve lesion, the elevated α-adrenergic activity associated with anxiety, life stress, and depression may contribute to RSD symptoms through its effect on sensitized receptors in the area of the lesion.

In addition to anxiety, life stress, and depression, certain personality factors may related to autonomic activity as well. For example, the type A behavior pattern, and especially the hostility component, has been found to be correlated with elevated SNS activity (17). Therefore, it is possible that certain personality patterns, through their association with elevated sympathetic outflow, may play a role in the development of RSD. Overall, the data presented above suggest the possibility that anxiety, life stress, depression, and specific personality styles may be involved in development of RSD through influences on sympathetic outflow, which in turn contributes to initial symptom development in individuals with nerve lesions.

CRITERIA FOR METHODOLOGICAL CRITICISMS

Studies of predisposing psychological factors in the etiology of RSD are maximally useful for making inferences to the extent that several methodological features are used. First, valid and reliable measures of psychological status must be used, with limited reliance on pure clinical judgment. Standardized psychodiagnostic criteria [i.e., Diagnostic and Statistical Manual of Mental Disorders (3rd ed., revised) (DSM III-R)] should also be explicitly used to allow comparisons across studies. A third methodological issue is the use of control groups, which can help address the question of whether individuals with nerve lesions who develop RSD are different psychologically from those with lesions who do not develop RSD. It is also important that studies have an adequate sample size. Standard psychological research practice suggests that 20 or more patients would be an acceptable sample size (18) and would help ensure adequate statistical power (19). These samples must also be unbiased; in particular, they should not be exclusively psychiatric referrals or treatment failures, or single cases reported because they are somehow unusual. A final methodological criterion relates to the temporality of the psychological factors relative to the onset of RSD. Ideally, prospective designs would be used to allow tests of causality. Given the difficulty of conducting prospective studies, retrospective designs may also be useful in that they provide more information regarding temporality than pure cross-sectional designs, although they have the disadvantage of recall bias. The literature below will be reviewed relative to these methodological criteria.

PSYCHOLOGICAL FACTORS IN ADULT RSD

One approach to investigating the etiological role of psychological factors in RSD is to assess the in-
idence of psychiatric disorders in RSD patients. Pak et al. (20) reviewed the records of 140 RSD patients seen at the Mayo Clinic to examine the physical and psychological findings in these patients. These records showed that 18% required formal psychiatric consultations during their treatment for RSD. These patients were given a variety of diagnoses, including anxiety, conversion reactions, and depression.

Other investigators have found an even greater incidence of psychiatric disorders in RSD patients. Poplawski et al. (8) reported that of 62 RSD patients, 31% of the overall group and 33% of the patients with long-term symptoms showed "overt psychiatric abnormalities" of an unspecified nature. It is unclear from the description presented by these investigators whether these abnormalities represented diagnosable psychiatric disorders. However, the fact that the investigators noted that all RSD patients in their study experienced at least some degree of anxiety suggests that the psychiatric abnormalities noted are more serious than minor anxiety reactions.

The studies presented above reflect only psychological status while being treated for RSD. These data do not indicate that psychiatric disturbance was present before the onset of RSD. Three studies have addressed the issue of temporality through the use of retrospective data-gathering procedures. For example, Horowitz (21) reported that of a group of 11 patients developing RSD subsequent to medical procedures, seven patients (64%) required psychiatric evaluations. All seven patients receiving psychiatric evaluations were diagnosed as experiencing clinical depression secondary to RSD. However, retrospective descriptions of the patients' psychological functioning indicated that six of these patients met the DSM III-R criteria for dependent personality disorder before developing RSD. In addition, these six patients were described premorbidly as having had a diminished ability to manage (unspecified) life stressors. One positive aspect of this study relative to other studies providing data regarding psychiatric disturbance in RSD patients is that the diagnoses were explicitly based on standardized criteria (i.e., DSM III-R). However, these data appear to have been obtained through retrospective self-report and may be biased by the effects of current psychological status.

Pak et al. (20) also obtained information regarding premorbid psychological functioning. They reported that 52 of 140 RSD patients (37%) had a history of psychiatric problems or emotional disturbances before the onset of RSD. The nature of these disturbances and the source of this information (psychiatric records or self-report) are not detailed. If this information were obtained primarily through self-report, the biased nature of retrospective data must be considered in drawing conclusions from these results. However, it is interesting to note that the premorbid prevalence of psychiatric disorders reported by Pak et al. (20) based on a large sample is consistent with the prevalence (31–35%) reported during treatment of RSD by Poplawski et al. (8), also a study using a relatively large sample. If the prevalence rate of psychiatric disorders in these two studies accurately reflects psychiatric status before onset of RSD, this prevalence is notably higher than the 19% overall prevalence rate for psychiatric dysfunction reported for the U.S. population (22).

Other investigators have attempted to assess premorbid psychological status in RSD patients by using behavioral measures as indicators of predisposing personality factors. Hemler et al. (23) studied the military records of 19 patients with RSD who were currently active in the military. These investigators operationalized personality predispositions to RSD as notations in the military record that set the patient apart from other active duty personnel in a negative way. Specific criteria used included reports of interpersonal conflicts with superiors, slow rate of promotion, and repeated sick calls for nonspecific complaints. Results indicated that the records of 63% of these patients were positive for one or more of the criteria listed above. Any conclusion that these results indicate psychological predisposing factors to RSD are based in part on the assumption that these criteria represent behavioral manifestations of distinct personality patterns. However, the validity of this assumption is questionable given the difficulty of inferring broad personality characteristics from a single behavioral criterion.

The studies reviewed above predominately used measures of psychological functioning (i.e., clinical interviews) whose reliability has been shown to be somewhat lower than might be assumed (24,25). Only one of the studies reported using standardized criteria for making diagnostic decisions (21). Unreliability in diagnostic decisions makes detection of possible patterns of psychological factors associated with RSD more difficult. One way to address these potential reliability problems is to use stan-
dardized psychological tests. Only four studies in the adult literature that we surveyed used standardized psychological measures, and these have all been published in the past 10 years.

The earliest of these studies, conducted by Subbarao and Stillwell (26), evaluated 125 cases of RSD. The Minnesota Multiphasic Personality Inventory (MMPI), a well-validated instrument designed to assess a number of areas of psychological functioning, was given to 45 of these patients. Of these 45 patients, 31% had abnormal scores on three scales reflecting somatic preoccupation (scale 1), depression (scale 2), and repression and hysteria (scale 3). Forty-two percent of the patients who completed the MMPI had abnormal scales 1 and 3 only, and 9% had abnormal scale 2 only. Normal profiles were found in 20% of these patients. The investigators noted that T scores of >69 or <51 were considered abnormal, but failed to identify whether the results they reported were due to high or low T scores. Treating scores of <51 T as abnormal, which implies that scores at the population mean (T = 50) are considered abnormal, is of questionable validity and may have biased the results of this study.

Very similar results for MMPI data were obtained by Grunert et al. (27). Twenty adult patients with RSD who were referred because of unsuccessful treatment completed the MMPI. Ninety percent of these patients had elevations on scales 1 and 3 only. One patient had elevations on seven of the 10 subscales, and one had a normal profile. Although the selection criteria were different, the similarity of findings in the two studies using the MMPI reviewed thus far is noteworthy.

Another study using the MMPI failed to detect any consistent pattern of RSD patient responses. Ladd et al. (28) reported on five RSD patients who received psychological testing including the short form of the MMPI (168 item) and the Family Environment Scale. The investigators reported that one of the patients had elevations on the MMPI on scales reflecting somatic preoccupation and depression (scales 1 and 2). However, these elevations were below the level usually interpreted as a cutoff for clinical dysfunction (T = 70). The other four patients had normal MMPI profiles. The Family Environment Scale, an instrument designed to assess several aspects of family functioning, indicated that three of the five patients perceived their families as low in cohesion and expressiveness, and one of these patients also indicated low levels of independence. High levels of family conflict were reported by another patient. The variety of responses represented in these data suggest no clear patterns of psychological or family functioning in RSD patients. However, the very small sample size for these psychological data severely limit their generalizability.

None of the preceding studies using the MMPI as a measure of psychological functioning has attempted to compare RSD patients’ test results with those of control patients. Only two of the studies reviewed used any comparison group whatsoever. Zucchini et al. (29) compared the MMPI profiles of 13 patients with RSD to 23 patients with nerve damage (brachial plexus lesions) without RSD. As with the Grunert et al. (27) and Subbarao and Stillwell (26) studies, the mean scores on scales 1, 2, and 3 obtained by RSD patients were found to be elevated. Although RSD patients’ scores on these scales were higher than the control group scores, these differences were not statistically significant because of the small sample size and large standard deviations. Nonetheless, the RSD patients were described as having significantly more “pathological” scale elevations than the controls, although this conclusion is somewhat limited by the use of an inappropriate cutoff (T > 60) for categorizing scale elevations as pathological. Clear interpretations of this study are also hindered by the fact that the RSD subjects were significantly older (48.7 vs. 24.8 years), and predominately female (11 of 13), whereas the control group was entirely male.

Haddox et al. (30) also compared psychometric data completed by RSD patients with those completed by controls. Twenty-one RSD patients and 21 radiculopathy patients completed the MMPI, the trait subscale of the State–Trait Anxiety Inventory (STAI; a measure of anxiety proneness), and the Dartmouth Pain Questionnaire (DPQ; a measure assessing the functional activity, pain behaviors, and self-perceptions of pain patients). Statistical analyses showed no significant differences on any of these measures between the two groups. The failure to find differences on the STAI between groups appears to be inconsistent with the theoretical model proposing that excessive anxiety can interact with nerve lesions to produce RSD symptoms. However, it should be noted that the STAI was completed after development of the chronic pain conditions, and therefore may not accurately reflect pre-morbid differences between the groups.

Overall, the studies using the MMPI suggest that
RSD patients tend to be depressed, somatically preoccupied, and use repression as a psychological defense. However, these same personality characteristics are often noted on the MMPI in all types of chronic pain syndromes (31); thus, these results indicate that RSD patients probably cannot be distinguished from general chronic pain patients on the MMPI. This assertion is also supported by the failure of the Haddox et al. (30) and Zucchini et al. (29) studies to distinguish between RSD and non-RSD pain patients. These results may indicate that RSD patients' personalities are no different than those of non-RSD pain patients. However, they may also indicate that the MMPI is not sufficiently sensitive to detect differences that may exist between the groups (13).

The results of one study regarding RSD in adults suggested that life stressors may be a factor in the development of RSD. Van Houdenhove (32) reported on 32 patients with RSD referred for psychiatric consultations because of depression, lack of cooperation with treatment, or suspected psychological problems. Clinical interviews showed that in 97% of these patients, a significant psychological stressor was associated temporally with the physically traumatic event that appeared to precipitate RSD. The psychological stressor in the majority of these patients was an affective loss (e.g., death, divorce, separation). Although consistent with the theoretical model proposed earlier, the generalizability of these data are severely limited by the fact that all of the RSD patients in this study were psychiatric referrals; thus, this sample is not representative of RSD patients as a whole.

Criticisms

Table 1 summarizes the findings and methodological strengths of the adult studies of psychological factors in RSD. The data regarding the involvement of psychiatric disorders and psychological stressors in the onset of adult RSD are limited by the methodological weaknesses throughout the literature. The most general criticism of this literature is that the dependent measures do not appear to have been well defined within studies. Most of the studies reviewed above use presence of abnormality determined in psychiatric interviews as the primary measure of psychological factors in RSD. The actual criteria used to define abnormality appears to vary from study to study, ranging from unspecified abnormalities to DSM III-R diagnoses. This would certainly explain the widely varying reports of prevalence of psychiatric disorders in RSD patients (18–64%).

Another weakness of these studies is that the populations evaluated sometimes appear biased. For example, Van Houdenhove (32) reported that all patients in his study were referred because of apparent psychiatric problems. Several of the other studies (e.g., 26) also imply that their populations might largely represent psychiatric referrals, although this is not clearly stated. Such an unrepresentative population would bias the results of these studies because psychiatric patients without RSD might also be expected to have abnormal scale elevations on the MMPI and report a large number of recent life stressors. The possibility that several of these studies used psychiatric populations would make the interpretation of the data regarding prevalence of psychiatric disorders in RSD even more problematic. Studies using samples referred because of treatment failure (e.g., 27) may be similarly biased.

The question of whether specific psychological factors predispose one to develop RSD is difficult to

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* Methodological criteria: a, valid/reliable measures of psychological status; b, explicit psychodiagnostic criteria; c, use of control groups; d, adequate sample size (20+ for adequate statistical comparisons); e, unbiased samples (i.e., not exclusively psychiatric referrals or treatment failures); f, prospective design; g, retrospective design.

* Cross-sectional data.

* Retrospective data.

* Based on MMPI: scale 1, somatic preoccupation; scale 2, depression; scale 3, hysteria.

* RSD patients were higher on MMPI scales 1–3, but comparisons with a control group were not statistically significant.
answer directly because only three studies in the adult literature attempted to evaluate psychological functioning before onset of RSD. There were no studies in the literature reviewed that used a prospective design. It also appears that the cross-sectional and retrospective designs used in these studies were not fully exploited. Only two studies (29,30) compared RSD patients with a control group; specifically, patients with radiculopathy or nerve lesions without RSD. Such control groups help address the question of whether RSD patients are more anxious or depressed than nerve-related pain patients without RSD.

Summary

It has been hypothesized that the presence of a psychiatric disorder, particularly anxiety or depression, may predispose one to develop RSD. Three of the more recent studies have found an elevated prevalence of psychiatric disorders in patients being treated for RSD. The only three studies that have attempted to assess the presence of psychiatric problems before onset of RSD have obtained results also suggesting a higher rate of psychiatric disorder in RSD patients. Thus, although this evidence is limited as a result of methodological flaws, the data are consistent with the hypothesis that anxiety and depression may contribute to development of RSD. The only study in the adult literature that assessed life stressors also reported a temporal relationship between the onset of RSD and life stress.

Psychometric information concerning personality characteristics associated with RSD suggests that, like other chronic pain patients, RSD patients are somatically preoccupied, depressed, and tend to use repression as a psychological defense mechanism. Because these data have not been obtained before onset of RSD, these personality characteristics cannot be described as etiological factors in RSD because they may simply be the result of having a chronically painful condition.

PSYCHOLOGICAL FACTORS IN CHILDHOOD RSD

Before 1977, only six cases of RSD in children had been reported in the literature (33). Since that time, however, a number of reports of childhood RSD have been published. As with the adult RSD literature, most of these studies are based on case reports and clinical interviews. Unlike the adult literature, however, many of the studies of RSD in children report personality characteristics rather than psychiatric diagnoses.

The first detailed study of RSD in a sample of children was conducted by Bernstein et al. (34). Of the 23 cases reviewed by these investigators, 57% had no known precipitating physical factors. Psychological consultation data were obtained on 17 of the 23 cases reviewed. Clinical interviews showed that these children could be characterized as having a tendency to assume excessive responsibility (85%), to have difficulty expressing anger or being assertive (90%), to be overly compliant with parental authority, and to have an indifference to the implications of the illness for future functioning (100%). Eighty-three percent of the families for whom psychological consultations were available had histories of overt parental conflict.

Similar observations were made by Sherry and Weisman (35). These investigators studied psychosocial factors in 21 families of children with RSD. Clinical interviews with these children suggested that they tended to be extremely compliant, unusually sensitive to the needs of others, and had difficulty expressing their own needs. Interviews with family members consistently showed inappropriate closeness between the parents and the children. This family enmeshment was exhibited both behaviorally (e.g., unwillingness to separate) and verbally (e.g., a mother and daughter describe themselves as being “like sisters”). All of these children were reported to have experienced multiple stressful events in the months preceding the onset of RSD. For example, in one patient who had been sexually abused, RSD developed 1 month before she was to testify against her assailant. Ten of these patients were experiencing stress in the form of academic problems. Another source of stress appeared to be family related; in nearly every family, parents were either divorced, separated, or experiencing significant conflict.

Psychometric data were also obtained by Sherry and Weisman (35). Scores on the Child Behavior Checklist completed by patients’ mothers indicated no major psychopathology, except for one patient scoring high on somatization. Ratings of family functioning obtained from parents using the Family Environment Scale suggested two patterns. In 71% of the families, cohesion, expression, and organization were high, with low to average levels of conflict; the remaining 29% of families were described as low in cohesion and high in conflict. Thus, unlike the interview information reported above, these re-
results suggest that family conflict was not a major factor in the histories of these children with RSD. This discrepancy might indicate that although there had been a high frequency of past family conflict, there was relatively little conflict at the time the Family Environment Scale was completed. This discrepancy might also reflect a desire to minimize ongoing conflict on psychological instruments. Because of these discrepancies, it is unclear how best to interpret these data.

A somewhat different characterization of children with RSD was reported by Silber and Majd (36). Interviews with 18 children and adolescents with RSD suggested that they could be described as having dependent personality traits, avoidance of responsibility, chronic complaining, insecurity, emotional lability, anxiety, and depression. Some of these children expressed a lack of concern for the consequences of RSD similar to that described by Bernstein et al. (34). Other patients seemed to "milkJ their disability, and thus, the RSD in these cases might be reinforced by secondary gain (i.e., the child was getting some positive benefit, such as attention, from having RSD). As was noted in the children interviewed by Sherry and Weisman (35), Silber and Majd (36) reported that the RSD seemed to be temporally associated with stress.

The possibility that ongoing stress may be related to treatment failures and relapse in children with RSD was suggested in a report by Dietz et al. (37). These investigators noted that in one of five childhood RSD patients who experienced recurrent RSD and continued pain despite standard treatment, a high level of current family stress was present. This stress was the result of leukemia in the patient's sibling, and was severe enough to precipitate family counseling.

Blau (38) also reported stress as a possible precipitator of RSD. The nine children and adolescents described in this study were reported to have experienced no identifiable physical trauma before onset of RSD. Interviews with these children showed that significant psychological stress and anxiety preceded RSD in 44% of these cases. For example, one adolescent's boyfriend had committed suicide recently. Another child was anxious about an impending move to a different state. Thus, in some cases, major life stressors were evident before onset of RSD, suggesting that they may have been directly related to its onset.

Rush et al. (39) also reported a case suggesting that stress preceded onset of RSD. They described a 9-year-old child who developed RSD in the hand and foot on two separate occasions, each time after a tooth extraction. In this case, no physical cause for the RSD was noted. Thus, it appears plausible that the anxiety accompanying the tooth extraction may have precipitated the RSD. Two other patients reported by these investigators had identifiable physical causes of their RSD, but were not experiencing any life stressors.

Several psychiatric disorders have been reported in children and adolescents with RSD. For example, anorexia nervosa was noted as preceding the onset of RSD in the case of a 14-year-old girl (40). Other serious psychological problems have been noted as well. Schiller (41) reported that of the seven adolescents with RSD who were described, the psychological histories of five indicated that they had experienced psychological problems, including school phobia and extreme academic stress, personality disorder, depression with suicidal ideation, and paranoia. These psychological problems were reported to be present before the development of RSD.

More commonly than the diverse disorders noted above, depression and life stress seem to be associated with RSD. Guntheroth et al. (42) reported the case of a 12-year-old boy with RSD who had no definite history of physical trauma, but was depressed and appeared to be facing excessive life stress. However, the report does not indicate whether the depression preceded the onset of RSD.

Another case more clearly indicates that depression and life stress preceded the development of RSD. Pilemer and Michelli (43) presented the case of a 16-year-old girl who was described by her parents as being significantly depressed before onset of RSD, and was experiencing several life stressors. These stressors included ongoing conflicts with male siblings and a physical attack by a stranger that occurred shortly before development of RSD symptoms. An MMPI and Beck Depression Inventory completed by the patient indicated depression, multiple somatic concerns, sexual identity confusion, and social inhibition. Although retrospective in nature, this particular case report clearly suggests that life stress and depression were present before the development of RSD.

Although the data are not uniformly supportive of the hypothesis that life stress may be a contributing factor in RSD, a recent clinical case in the second author's (C.R.C.) practice illustrates the potential connection. In this case, a 9-year-old girl inexplica-
bly began to lose functioning in her left leg, and reported that the foot was too painful to touch or to use. Just before the onset of RSD, the patient’s parents had informed the child of their impending separation/divorce. The temporal relationship between the onset of the RSD and the stress seemed quite striking. Interestingly, the resolution of the RSD through intensive biofeedback training, physical therapy, and epidural sympathetic blockade occurred without a significant reduction in the conflicts associated with the estranged husband-wife relationship.

Criticisms
The literature concerning RSD in children has many of the same weaknesses as the adult literature. The use of qualitative descriptions of personality traits derived from clinical interviews, although informative, can be based on variable criteria and can be difficult to integrate across studies. Thus, clear psychological profiles associated with RSD, if any, are not easy to identify. One serious shortcoming is the lack of standardized measures of personality functioning and ongoing emotional states in the children in the studies reviewed. As with the adult literature, it appears that many of the children with RSD for whom psychological information is available are those who may have been referred because of psychological problems or poor treatment response. The lack of adequate studies assessing psychological functioning before onset of RSD in children makes it difficult to draw conclusions regarding psychological factors in the development of RSD. Finally, the small sample size typical of these studies, especially the reliance on single-subject case studies, limits the generalizability of many of the results reported.

Summary
The psychological factors found to be associated with RSD in children are presented in Table 2. Definitive conclusions regarding the role of environmental or psychological factors in the development of childhood RSD are not possible at this time due to the weaknesses in the existing literature. However, the results of our review are consistent with the theoretical model which proposes that anxiety, life stress, and depression may serve as etiological factors in RSD. These three factors were mentioned as contributing to RSD in 10 of the 11 studies of childhood RSD reviewed. Specific life stressors mentioned ranged from academic difficulties to physical abuse and assault, as well as family and parental conflict. However, conclusions regarding the role of anxiety, life stressors, and depression in RSD must be tempered by the cross-sectional and retrospective nature of these data.

Four of the 11 studies reviewed specifically note that there tended to be no history of physical trauma to account for the RSD. Although this suggests that psychological factors might therefore be responsible, the possibility that there was a physical trauma that went unreported or undetected cannot be ruled out. This latter possibility seems likely given the current understanding of the physiological basis of RSD. Life stress, anxiety, and depression, if they are involved in the development of RSD, are most likely contributing to the SNS component of RSD that may develop after physical injury.

The evidence for psychological traits as etiological factors in childhood RSD is limited. Generalizations across these studies are difficult because of the qualitative nature of these data. If certain personality traits are associated with RSD in children, these might include excessive compliance with the wishes of others, lack of assertiveness, and a tendency to assume excessive responsibility.

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**TABLE 2. Psychological factors associated with RSD in children**

<table>
<thead>
<tr>
<th>Reference</th>
<th>Psychological factors</th>
<th>Methodological criteria met*</th>
</tr>
</thead>
<tbody>
<tr>
<td>34</td>
<td>Excessive responsibility, difficulty with anger expression and assertiveness, overcompliance, parental conflict</td>
<td>d.g</td>
</tr>
<tr>
<td>35</td>
<td>Overcompliance, difficulty expressing own needs, overinvolvement, life stressors/anger</td>
<td>d.e.g</td>
</tr>
<tr>
<td>36</td>
<td>Normal child behavior, families reported high cohesion, emotional expression, and organization</td>
<td>a.d.e</td>
</tr>
<tr>
<td>37</td>
<td>Anxiety, depression, dependency, emotionally labile, insecure, avoid responsibility</td>
<td>e.g</td>
</tr>
<tr>
<td>38</td>
<td>Life stressors/anxiety</td>
<td>g.g</td>
</tr>
<tr>
<td>39</td>
<td>Life stressors/anxiety (tooth extraction)</td>
<td>g.h</td>
</tr>
<tr>
<td>40</td>
<td>Anorexia nervosa</td>
<td>g.g</td>
</tr>
<tr>
<td>41</td>
<td>School phobia, depression, paranoia, personality disorder, life stressors/anxiety</td>
<td>e.g</td>
</tr>
<tr>
<td>42</td>
<td>Depression, life stressors/anxiety</td>
<td>e.g</td>
</tr>
<tr>
<td>43</td>
<td>Depression, somatic preoccupation, anxiety, life stressors/anxiety, social inhibition</td>
<td>g.a</td>
</tr>
</tbody>
</table>

* See Table 1 footnote.
* Based on MMPI: scale 1, somatic preoccupation; scale 2, depression.
GENERAL DISCUSSION

The disparity between the seemingly minor trauma believed to have initiated RSD and the extreme pain experienced by the patient can cause health professionals to conclude that RSD patients are malingered, neurotic, or emotionally unstable (9). Psychological factors have been postulated as a cause of RSD in part because of a lack of obvious physical causes for the degree of pain reported. Recent research suggests that physiological factors can account for the degree of pain observed through the interactive effects of nerve lesions and SNS sensitization of mechanoreceptors or nociceptors (3, 7, 14). However, this physiological understanding of the etiology of RSD does not eliminate a role for psychological factors in the development of the disorder. It is theorized that anxiety, life stress, and depression may contribute to the development of RSD through their effects on α-adrenergic activity (16). If psychological factors are indeed etiologically significant in RSD, they are most likely contributory, but not necessary factors for development of the disorder. As has been suggested in the literature (1, 8), it is possible that in some cases psychological factors may make certain individuals susceptible to developing RSD, whereas in other cases, the psychological factors associated with RSD patients are a result of having a chronically painful condition.

With the current lack of well-controlled and systematic studies, it is difficult to draw conclusions regarding the role of psychological factors in the etiology of RSD. Our review indicates that depression, anxiety, and life stress (primarily in children) are psychological factors frequently reported to be associated with RSD. Of the 20 published studies of RSD patients reviewed, 15 note the presence of one or more of these three factors. These data are consistent with the theoretical model proposed earlier.

Although our review indicates that depression, anxiety, and life stress may often be present in individuals with RSD, the question of whether these psychological factors preceded and therefore were etiologically related to RSD cannot be answered due to the methodological weaknesses of existing studies. There is a small body of retrospective evidence suggesting that premorbid presence of these psychological factors in adults and children may be associated with development of RSD. However, these data often lack standardized criteria for identifying psychopathology. The results of some of these studies also may be biased because they were based on psychiatric referrals or treatment failures. It should be noted that these studies generally were not designed specifically to test hypotheses regarding psychological factors in RSD; therefore, the methodological weaknesses noted are not surprising. Until well-designed prospective studies are conducted, the hypothesized association between psychological factors and development of RSD cannot be confirmed.

If psychological factors do not play a role in the etiology of RSD, they clearly do play a role in determining the severity of the experienced pain and disability. Poplawski et al. (8) noted that all RSD patients, even those without psychiatric abnormalities, experienced anxiety about control of their symptoms and their prognosis. Anxiety and depression can contribute to increased sensations of pain (44–46), as well as increased SNS activity (16), which contributes to the pain of RSD. Thus, even if RSD develops without the contribution of psychological factors, the degree to which pain is reported will be a function of the various psychological factors operating in the patient’s life.

In addition to the emotional state of the patient directly affecting the level of pain, psychological factors also might be involved in the maintenance of the disorder through behavioral changes. Some investigators have argued that the persistent pain of RSD may result largely from the restriction of mobility that comes from trying to protect the affected areas (47). When family members or co-workers take over the responsibilities of the RSD patient, restricted mobility continues, thus prolonging the disorder. Other investigators have hypothesized that given the excessive compliance and responsibility often exhibited by children with RSD, the presence of RSD may serve a functional role. For example, RSD may offer children a means of decreasing home responsibilities without having to confront parents (34, 35). This hypothesized role of secondary gain in maintaining RSD is consistent with general theories of chronic pain (e.g., 48).

The presence of anxiety, depression, or high levels of life stress may contribute to the percentage of RSD cases that remain resistant to treatment or eventually relapse, although this hypothesis remains untested. For example, Pak et al. (20) reported that 20% of 75 patients receiving standard RSD treatment (e.g., sympathetic blocks, physical therapy) failed to improve. Similarly, Subbarao and Stillwell (26) indicated that of 77 patients for whom
follow-up data were available, 21% had continued pain and stiffness despite standard treatment. Standard medical and physical therapy approaches to RSD are effective in a large percentage of cases. However, a percentage of RSD patients relapse or fail to improve despite sympathetic blockade and physical therapy (20,26). The theory proposed above suggests that at least in some of these patients, psychological factors (e.g., excessive anxiety) should be considered as a possible factor maintaining the RSD, through effects on sympathetic outflow, despite medical and physical therapy treatment.

One standard approach to treating RSD relapse is surgical sympathectomy. A less invasive alternative may be the use of psychological treatments (e.g., relaxation and biofeedback) focused on teaching the patient to reduce SNS activity (27). Such an approach, although it has not been sufficiently evaluated, may improve treatment outcome for some patients without the potential negative side effects of surgical sympathectomy and, at the very least, is worthy of careful examination.

CONCLUSIONS AND DIRECTIONS FOR FUTURE RESEARCH

The question of whether psychological factors predispose certain individuals to RSD cannot be answered at this time due to the methodological weaknesses of the literature. However, the literature reviewed is consistent with a theoretical model in which the premorbid presence of depression, anxiety, and life stress may result in sympathetic hyperaerial, which can contribute to the development and/or maintenance of RSD. Confirmation of this hypothesis must await future research. Even in the absence of definitive conclusions, it is clear that psychological factors must be addressed in the treatment of RSD patients. As pointed out in one of the early reports on RSD, "the early relief of the continuous, intractable pain and the proper reassurance of the patient may prevent the late complications" (10). Thus, medical treatment should include a multidisciplinary approach including medical staff, psychologists, and physical therapists in order to resolve the symptoms of RSD in a timely manner. If future research identifies specific psychological risk factors for development of RSD, it may be appropriate to provide brief preventive psychological assistance (e.g., relaxation training) for at-risk individuals who have sustained physical trauma that may be commonly associated with development of RSD.

Given the potential therapeutic significance of the confirmation of a role for psychological factors in the development of RSD, the need for further research cannot be overemphasized. Future RSD research would benefit greatly from greater attention to methodological rigor. For example, more systematic use of well-validated psychological measures assessing the areas of personality functioning, emotional state, trait anxiety, and life stress would be helpful for gaining more detailed knowledge of psychological factors in RSD. Where clinical interviews are used to make psychiatric diagnoses, standard structured interview protocols should be used. Review of the current literature indicates that more frequent use of appropriate control groups, especially from other medical populations, would also add to the interpretation of the conclusions.

Although a number of research options are available (e.g., better designed cross-sectional or retrospective studies), only a prospective study will specifically answer the question of whether psychological factors are involved in the etiology of RSD. However, one difficulty with this research strategy is obtaining adequate samples of psychological information on RSD patients before RSD has developed. Although accurate estimates of the incidence of RSD are difficult to make due to varying diagnostic criteria, the incidence of causalgia after peripheral nerve injury of 1\%-5\% (1) may be taken as an estimate of RSD incidence after traumatic injury. Given this low base rate, prospective research may be facilitated through the use of coordinated multisite data collection focused on susceptible populations. The current literature suggests potential populations for further study. A review of 125 cases of upper extremity RSD (26) indicated that 45\% of these cases followed fractures of the wrist, with an additional 6\% following hand surgery. Thus, while not the only available data-collection strategy, data gathering in multiple orthopedic clinics may provide an opportunity to obtain samples adequate for prospective analysis.

One possible research strategy would be to systematically collect standardized psychological data (assessing personality, psychopathology, emotional state, and life stress) on patients who have recently sustained acute traumatic injuries while they are at the orthopedic clinic. The statistical power of this research can be enhanced through use of standardized measures such as the Daily Hassles Scale (49).
as a measure of life stress, the Beck Depression Inventory (50), and the STAI (51), the latter which has been found to correlate with alpha-adrenergic receptor activity (16). Personality functioning could be assessed in both adults and adolescents through use of the MMPI-2, although this instrument may not be sufficiently sensitive to the personality characteristics of interest (13, 30). An alternative measure of personality functioning is the NEO Personality Inventory (52), which has also been well validated and has the advantage of being much shorter than the MMPI-2. Once a sufficient sample size has been obtained (i.e., at least 20 patients in the sample have developed RSD), personality profiles, depressive symptomatology, anxiety proneness, and life stress of individuals who developed RSD could then be compared with those who did not. Obtaining these psychological measures before long periods of chronic pain have occurred minimizes the likelihood that the RSD itself may have caused the psychological factors associated with RSD. Until such well-defined prospective studies are conducted, the question of whether psychological factors predispose certain individuals to the development of RSD must remain unanswered.

Despite the need for prospective studies, the costs and benefits of conducting such studies must be considered. Given the low base rate of RSD, it is likely that collection of sufficient data to allow statistically powerful tests of the hypotheses would require an extended period of time. For example, assuming a base rate for RSD of 5% in the design described above, data on 400 traumatically injured patients would have to be obtained in order to collect the minimum adequate sample of RSD patients. In addition to this potential difficulty, it is likely that unless subject recruitment is consistently and diligently performed, the loss of potential subjects may significantly bias the results of these prospective studies. Thus, the cost in time and money of a well-designed prospective study may be high, and it will be up to researchers to determine whether the costs offset the potential benefits of this type of study.

The suggested prospective research has implications for clinical practice. Empirical confirmation of psychological factors in the etiology of RSD would point to the need for more regular adjunctive psychotherapeutic care in the treatment of RSD. If psychological factors are clearly shown not to be involved in the etiology of RSD, automatic assumptions that all RSD patients are psychiatrically disturbed should be revised to protect such patients from the negative effects such an assumption may engender (e.g., 53).

Acknowledgment: This work was supported in part by Grant MH15730 from the National Institute of Mental Health. We thank Marianne Brady for her contribution to this project, and Dr. Peter R. Wilson and several anonymous reviewers for their constructive comments on earlier versions of the manuscript.

REFERENCES
REFLEX SYMPATHETIC DYSTROPHY


