Psychiatric approach in the treatment of reflex sympathetic dystrophy in an adolescent girl: a case report

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Reflex sympathetic dystrophy (RSD) is an unusual diagnosis in the pediatric age group. It is a syndrome characterized by pain in one or more extremities with a significant morbidity in childhood. Patients with RSD have frequently undergone many unnecessary investigations such that the diagnosis and treatment may be considerably delayed. The pathophysiology remains unclear; however, a number of psychological problems were frequently suggested to play a role in this disorder. We describe a 13-year-old girl diagnosed as pediatric RSD who was admitted to a child and adolescent psychiatry unit with a history of severe pain in the right hand, increasing disability and symptoms of nervousness and withdrawal from social activities. In this report, we discuss psychogenic factors underlying the disorder of an adolescent girl and psychiatric approach as a part of a multimodal treatment of pediatric RSD.

Key words: reflex sympathetic dystrophy, treatment, psychiatric approach.

Although more commonly seen among adults, reflex sympathetic dystrophy (RDS) may also occur in children and adolescents. However, it has been suggested recently that this syndrome could be underestimated and misdiagnosed in the pediatric age group[1]. Typically, the condition presents with chronic, painful swelling in the extremity, decreased skin temperature, cyanosis, delayed capillary refill, and limitation of functioning.

This disorder is a form of local causalgia, usually involving a hand or food but not corresponding to the anatomic distribution of a peripheral nerve. The pain is debilitating and is exacerbated by the movement of an associated joint. Though no objective signs of arthritis are seen, immobilization provides some relief. The most common preceding event is local trauma[2].

The pathophysiology of RSD is still undetermined, but several pathogenetic theories have been proposed, such as dysregulation of the sympathetic nervous system or an inflammatory response to injury[3]. In general, there appears to be an association of RSD with psychosocial stressors. Psychological mediators have been proposed and outlined in case series[4-7]. Also, some researchers have reported that most of the patients had some psychological disturbances like histrionic reactions and nonorganic somatic complaints[6].

Considering the strong effects of psychological factors on the etiology of RSD, psychiatry has an important role in the prognosis of the illness as a part of the multidisciplinary approach. Thereby, we report the history and the administered therapy of a girl patient with a diagnosis of RSD who was referred to the Department of Child and Adolescent Psychiatry.

Case Report

TB, a 13-year-old girl who was unable to use her severely aching right hand with the additional symptoms of nervousness and withdrawal from social activities, was referred to the outpatient clinic of Child and Adolescent Psychiatry. She reported that the pain began suddenly during the cleaning of the house eight

months before her admittance. She underwent an operation for compartment syndrome by the orthopedic surgeons, but the pain did not improve and progressively increased. She was reoperated after two months followed by an intense physiotherapy program, after which she was able to partially move her fingers, but she reported no improvement in the pain. Although she had received an intermittent course of physiotherapy until the time of her admittance, she was unable to move her hand and forearm. A number of non-steroidal anti-inflammatory drugs (NSAIDs), tricyclic antidepressant (TCA), corticosteroid and beta adrenergic blockers had been administered. The atrophic changes started in her hand and fingers. As tiny touches to her hand produced severe pain, the nails of the right hand could not be cut. Neurologic examination, computerized tomography, electromyogram (EMG) and laboratory results revealed normal findings, after which any comorbidity that could account for the degree of pain and dysfunction was excluded. Based on the criteria[8] that symptoms started after a noxious event without an apparent nerve lesion, that pain was disproportionate to the inciting event, and that trophic changes (nail growth, brittle nails and tissue atrophy) developed after the event, the diagnosis of RSD was considered.

TB was born by normal vaginal delivery as a second child of a nucleus family with two children. It was a planned pregnancy. No problems were reported in prenatal, natal, or postnatal periods, and she met all of her milestones at normal ages. She attended primary school until the third grade and then discontinued school following the earthquake. She had numerous responsibilities at home after quitting school. Her mother, a 38-year-old housewife, graduated from primary school and her father was a 41-year-old worker. The decisions in the house were generally made by the mother. The father was a calm and passive man. The brother of TB graduated from secondary school and was working as a musician. He had a history of acute rheumatic fever and had not been given any responsibilities, especially by his father, because of his illness. TB assumed the responsibilities which her brother would normally have done.

The patient was hiding her hand in the abducted position during the first interview in the outpatient clinic. It was observed that she did not want to be alone and that she was quiet and nervous. She was overreacting in response to the questions about her hand. She was offering short answers and generally pretending not to understand the questions, i.e. acting immaturely. The psychiatric evaluation and the psychologic investigations revealed that she had intensive depressive symptoms. A course of fluoxetine treatment at a dose of 20 mg/day and individual psychotherapy were started. Following the psychotherapy, she was hospitalized in the ward of the Physical Therapy and Rehabilitation Department. She refused all the treatment modalities because of the pain in her hand and argued with the physiotherapists, accusing them of hurting her at all times. In the second week of her hospitalization, she had fainting attacks while being transported for the physiotherapy or during the therapy. The neurologic examination and the EEG findings were normal. She was doing very well doing and had a joyful demeanor at times outside of therapy. Although she underwent a sympathetic nerve blockage procedure at the department of anesthesia, she reported no regression in her pain but instead a progressive increase. The blockage was removed and she was discharged after 20 days of hospitalization.

In addition to the individual therapy, her family participated in the psychotherapy during the follow-up period. It was observed that TB continued to have an immature behavior that was supported by her parents. Despite the normal pediatric examination and laboratory findings within normal limits, she had severe abdominal pain. The dominant behavior of the mother over her passive husband, who seemed to await confirmation, was strikingly apparent. The brother tended to be quiet and was not interested in the environment. The studies performed with the family revealed the discordance between the mother and the father and the symbiotic relationship developed by the mother between her own parents and her children. It was also observed that the role and responsibilities of the mother had been transferred to our patient a long time previously. It was interesting that one reason for the ineffectiveness of the therapy was that the mother had frequently sought new treatment modalities since the beginning of the illness. The decreased frequency of discussions between the parents, TB's decreased responsibilities,

and the mother's increased absences from the house were suggested to be secondary benefits caused by the illness.

Both individual and family therapies were based on an eclectic psychiatric approach. During the sessions, efforts were made to resolve the symbiotic relationship of the family, especially of the mother. Efforts were given towards improving TB's immature behavior and encouraging her individualization, and her family was encouraged to treat her age appropriately. It was attempted to establish a redistribution of the roles and responsibilities at home.

At the end of the one-year follow-up period, the patient could touch her hand and arm. Her nails could be cut and she had started hand exercises with a small ball. She slowly returned to normal daily activities and started to use her hand. The physiotherapy programs and the interviews at our clinic are continuing.

Discussion

A patient with RSD who was referred to the outpatient clinic of the Child and Adolescent Psychiatry Department is presented in this case report. RSD is a syndrome with poorly defined diagnostic criteria[5,9] characterized by pain and vasomotor disturbances. The first symptom is pain at the site of injury which progresses either proximally or distally regardless of a dermatomal distribution or anatomic landmarks. Generalized swelling and vasomotor disturbances of the limb are seen in 80% of the affected children. The children report an intense pain described as burning or aching. Pain is exacerbated by movement or dependence, causing the arm to be held in a position of abduction and internal rotation as if it were swaddled to the body[8]. RSD usually starts with an acute stage, followed by a dystrophic period and finally by atrophy. The mean age at onset in children is 11, and girls are affected more then boys [5,10].

Although 200 cases were reported up to 1995[11], the authors claimed that the disorder was more prevalent than generally thought[6]. It has been suggested already that RSD is poorly recognized in the pediatric age group and that misdiagnosis may still occur. In a large study of 70 patients, the interval from the onset of symptoms to the time of the diagnosis averaged 12 months[10]. Dietz et al.[12] reviewed 80 patients from different reports and described a mean delay to diagnosis of six months despite the performance of numerous invasive tests during this period. Cleary et al.[13] noted a significant delay in diagnosis in many children, with a median time to diagnosis of 12 weeks.

Our patient had also undergone several diagnostic procedures and two operations. She was diagnosed by means of several invasive techniques and the treatment was started afterwards. In several cases, a history of trauma was considered the triggering event; however, no evidence of trauma was found in many children [8,12]. Our patient had a history of trauma coinciding with the beginning of the disorder.

Psychological problems play a major role in this disorder. Sherry and Weisman[4] analyzed the psychosocial factors in 21 families of children with RSD, by means of interviews and standardized psychologic tests, and pointed out that RSD is frequently a disease related to stress. Cimaz et al.[6] reported that most of the patients had some psychological disturbances like a histrionic reaction, nonorganic abdominal pain and vomiting and clustering of somatic pains. The history of our patient together with her conversive fainting attacks, abdominal pain and immature behavior seemed to hint at the underlying psychological problems. Parental participation in the treatment, re-identification of the roles in the family, resolution of the symbiotic relationship, establishment of separation-individualization of the patient and the disappearance of the accompanying depressive symptoms accelerated the patient's physiotherapy program and the duration of the improvement. As for prognosis, it seems that the condition could be more benign in children than in adults[14], even though there are cases in the literature which are severe and resistant to treatment compared to the adult patients[11,15,7,16]. The response to treatment also depends on the duration of symptoms, because nonresponders have usually been patients in whom diagnosis has been delayed or those with severe and long-lasting underlying psychologic problems. Radler et al.[17] reported that the treatment of RSD with pharmacologic agent in combination with psychological counselling was a safe and effective treatment regimen. Additional psychological counselling helps

patients and their parents to develop coping strategies which may help to avoid relapses.

In conclusion, we described herein the underlying psychogenic causes and the treatment process with a multimodal approach in a girl patient with RSD. The individual and family psychotherapeutic treatment with a multidisciplinary approach has important and positive effects on the prognosis of RSD, which has been included under somatoform disorders in the textbook of psychiatry[18].

Chances for full recovery seem to be better for patients treated early, while those with delayed diagnosis and prolonged symptoms often have persistent disease despite aggressive treatment[1]. Early attention to such patients may avoid unnecessary organic work-ups; promote early diagnoses and timely psychological, medical and pharmacological treatments; prevent negative consequences; and decrease morbidity.

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