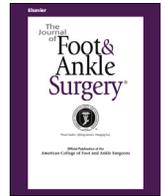




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Original Research

Incidence of Complex Regional Pain Syndrome after Foot and Ankle Surgery

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ABSTRACT

Complex regional pain syndrome (CRPS) is an uncommon complication of orthopedic surgery, and few investigators have considered the incidence in foot and ankle surgery. In the present retrospective cohort study of 390 patients who had undergone elective foot and/or ankle surgery in our department from January to December 2009, the incidence of postoperative CRPS was calculated and explanatory variables were analyzed. A total of 17 patients (4.36%) were identified as meeting the International Association for the Study of Pain criteria for the diagnosis of CRPS. Of the 17 patients with CRPS, the mean age was 47.2 ± 9.7 years, and 14 (82.35%) were female. All the operations were elective, and 9 (52.94%) involved the forefoot, 3 (17.65%) the hindfoot, 3 (17.65%) the ankle, and 2 (11.76%) the midfoot. Twelve patients (70.59%) had new-onset CRPS after a primary procedure, and 5 (29.41%) had developed CRPS after multiple surgeries. Three patients (17.65%) had documented nerve damage intraoperatively and thus developed new-onset CRPS type 2. Blood test results were available for 14 patients (82.35%) at a minimum of 3 months postoperatively, and none had elevated inflammatory markers. Five of the patients (29.41%) were smokers, and 8 (47.06%) had had a pre-existing diagnosis of anxiety and/or depression. From our findings, we recommend that middle-age females and those with a history of anxiety or depression, who will undergo elective foot surgery, should be counseled regarding the risk of developing CRPS during the consent process. We recommend similar studies be undertaken in other orthopedic units, and we currently are collecting data from other orthopedic departments within Scotland.

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Complex regional pain syndrome (CRPS) was first described by Mitchell (1) in 1864 as a “burning pain” experienced by injured soldiers in a Civil War. Currently, it is known as a neuropathic pain syndrome that can occur postoperatively as a complication of surgery. The pathophysiology was relatively poorly understood in the past, with no definitive treatment available, and the symptoms can be debilitating for patients (2). The diagnosis of CRPS is made clinically, and no diagnostic tests are available. Little information is available in published studies regarding the types of patients who develop CRPS specifically in relation to foot and ankle surgery. Our main aim, therefore, was to determine the incidence of CRPS in patients who had

undergone foot and/or ankle surgery and to establish the patient factors associated with its development.

Patients and Methods

The present study was a retrospective cohort study of 390 patients who had undergone foot and/or ankle surgery at the Western Infirmary in Glasgow, Scotland, from January to December 2009. All 390 patients had undergone surgery on an elective basis by the same foot and ankle team consisting of 2 primary surgeons (J.S.M. and R.M.). They had either documented “CRPS” (either CRPS Type I, reflex sympathetic dystrophy syndrome, or CRPS Type II, causalgia) within their clinical notes or documented signs and symptoms fulfilling the International Association for the Study of Pain diagnostic criteria. These include the presence of an initiating noxious event or a cause of immobilization; continuing pain, allodynia, or hyperalgesia in which the pain is disproportionate to any known inciting event; and evidence at some point of edema, changes in skin blood flow, and/or abnormal sudomotor activity in the region of pain. The diagnosis can be excluded by the existence of other conditions that would otherwise account for the degree of pain and dysfunction (3). The information was collated manually by 2 physicians (A.H.L. and A.G.), who reviewed every patient who had undergone surgery by the foot and ankle team and extracted details from the medical records or Bluesp

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(Bluespeir International, Droitwich, England), a computerized database containing patients' preoperative, intraoperative, and postoperative clinical notes. Where possible, the patient case notes were reviewed, because these usually contained more comprehensive information. The patients were followed for a minimum of 1 year.

Our outcome of interest was the identification of CRPS in the postoperative period of observation, and the exposures (independent variables) of interest included age, gender, the onset of CRPS (pre-existing or new in the postoperative phase), type of CRPS (diagnosis type 1 vs type 2), anatomic site of surgery, clinical signs, and inflammatory marker levels. Our aim in this retrospective cohort study was simply to calculate the incidence of CRPS and to observe number of selected demographic variables associated with the diagnosis.

Results

A total of 390 patients were identified. Of these patients, 17 (4.36%) had a diagnosis of CRPS postoperatively. Their mean age was 47.2 ± 9.7 (range 27 to 61) years. Females were more likely to be affected ($n = 14$, 82.35%). Twelve patients (70.59%) acquired new-onset CRPS, 2 (11.76%), who had undergone previous foot and ankle surgery, had brief resolution, and 3 (17.65%), who had undergone previous surgeries, had continuing signs and symptoms of CRPS. Fourteen patients (82.35%) had type 1 CRPS, and 3 (17.65%) had CRPS type 2. All patients had pain, and nearly all ($n = 16$, 94.12%) had skin changes. Of all the patients who developed CRPS, 3 (17.65%) had undergone surgery at the ankle, 3 (17.65%) at the hindfoot, 2 (11.76%) at midfoot, and 9 (52.94%) at the forefoot. Regarding the relevant medical history of the patients with CRPS, 8 (47.06%) had a medical history of anxiety or depression, and 5 (29.41%) were smokers. Of the 17 patients with confirmed CRPS, 14 (82.35%) had inflammatory marker test results available, and their white blood cell count and C-reactive protein levels were all normal.

Discussion

CRPS is an uncommon complication in foot and ankle surgery, and it is characterized by severe pain, skin changes, and swelling (4). The natural history has been characterized by chronicity and relapses that can result in disability. CRPS has had many synonyms, which has led the International Association for the Study of Pain to define 2 types of CRPS (3). Type 1 (formerly known as *reflex sympathetic dystrophy syndrome*) results from a noxious event or a cause of immobilization. Patients experience continuing pain, allodynia, or hyperalgesia disproportionate to the initiating stimulus. Edema, changes in skin blood flow, or abnormal sudomotor activity will be present. However, other diagnoses should be excluded. Type 2, formerly termed *causalgia*, refers to the presence of pain, allodynia, or hyperalgesia specifically after a nerve injury but not specifically in the distribution of that nerve and includes the features of type 1 (5).

The pathophysiology includes different mechanisms of motor, sensory, and autonomic dysfunction and is not entirely clear. However, no evidence has shown that the pathophysiology differs for the different CRPS types. The main hypotheses have included peripheral and central sensitization, inflammatory changes, altered sympathetic and catecholaminergic interaction, reduced somatosensory representation in the brain cortex, genetic components, and psychophysiological components (2,6).

Understanding the pathophysiology of CRPS can often be difficult and, therefore, identifying it in clinical practice and ultimately treating CRPS will be equally difficult. Recognizing the risk factors or patient characteristics predictive of CRPS could be key to prevention and treatment.

Of our patient population with CRPS, 82% were female. Other evidence in published studies has indicated that it is often females who are affected. A 10-year population-based study of the incidence of CRPS type 1 also found females to be 4 times more likely than males to develop CRPS, with an average age of onset at 46 years, similar to the age of onset at 47 years in our population (7). Another study reported

that predominantly white, middle-age females were at the greatest risk (8). They also reported that CRPS type 1 accounted for 96.9% of their cases (8). Anderson and Fallet (9) had similar findings specifically in their foot and ankle patients. Females were mainly affected, the most common manifestation was type 1, and the most common causative foot surgery was neuroma excision (9). Moreover, McNerney (10) established that the most basic of elective surgical procedures of the lower limb can result in CRPS and, therefore, all patients must be made aware of the risk.

A modest number of studies have attempted to identify patient characteristics indicative of developing CRPS but few specifically in the foot and ankle. After studying our cohort and their medical history, we found that almost one half of the foot and ankle candidates who developed CRPS had had anxiety or a depressive history. Hemler et al (11) also reported that patients with a premorbid history of depression or anxiety were more likely to develop CRPS.

Only 30% of our patients with CRPS in the foot or ankle were smokers. However, other studies have found that smoking increases the risk of developing CRPS. Howard et al (12) reported that 68% of their patient cohort were smokers and believed the reason was the possible increased likelihood of peripheral vasospasm. Their study (12) was not specific to foot and ankle patients, and our results contradict their findings.

Few treatments have been effective because of a historically poor understanding of the mechanisms underlying the disorder. However, we have identified middle-age females and those a medical history of depression or anxiety to be at risk of developing CRPS. Such patients should be identified preoperatively and counseled about the potential complication of CRPS (13).

Our study has demonstrated important clinical features associated with CRPS. All our patients had pain, and most had erythema or edema surrounding the foot or ankle. These features were supported by 1 study that stated that differences in limb skin temperature could aid in the diagnosis and that this was dependent on spontaneous sympathetic activity (3). Because most of our patients had normal inflammatory marker levels, this would suggest that an inflammatory component is not associated with CRPS development. We also did not find radiographic features specific to CRPS, which was also reported by Bacchini et al (14), who demonstrated that the diagnosis of CRPS after foot and ankle surgery is solely clinical.

Patients with CRPS experience intense pain but also significant functional disability and psychological distress (15). Early detection and treatment will help prevent long-term disability and recognizing those most at risk will aid in detection (16,17). It is important to understand that the treatment should be multidisciplinary, with the aim of controlling symptoms and restoring function. This includes pharmacologic treatment, nerve blocks, psychotherapy, and physiotherapy. Patients should be closely followed up to ensure progress (18,19). One recent retrospective study by Dellon et al (20) identified 30 patients with CRPS type 1 and followed up 13 who were treated operatively, including joint denervation, neuroma resection, muscle implantation, and neurolysis. Of these patients, 55% had excellent results, with either a reduction in pain medication or a restoration of function, 30% had good results, and 15% did not experience CRPS symptom resolution (20).

The limitations of the present study included that all patients were from 1 geographic location at 1 hospital and had undergone elective surgery. Other limitations included the inherent problems with retrospective studies and that we relied on the documentation of CRPS from the clinical notes or signs and symptoms suggestive of CRPS. Therefore, the actual number of patients with CRPS might have been under- or overdiagnosed. Future research will require large samples of foot and ankle patients who have developed CRPS to be examined using experimental designs that reflect the multifactorial nature of CRPS. This would aid in making causal links and developing potential therapies, reducing patient suffering and healthcare costs (18).

We have clearly identified that middle-age females and those with a history of anxiety or depression have a greater risk of developing CRPS after foot and ankle surgery. We should, therefore, ensure that such patients are aware of the risks and other potential management options, because the chronic pain could be more debilitating than the reason they were planning to undergo elective surgery.

In conclusion, females and those with a history of anxiety or depression undergoing elective foot and ankle surgery should be counseled regarding the risks of developing CRPS during the consent process.

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