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CASE REPORT

CRPS I following artificial disc surgery: case report and review of the literature

S. M. Knoeller · M. Ehmer · B. Kleinmann · T. Wolter

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Abstract We report a case of type 1 complex regional pain syndrome (CRPS I) of the left leg following the implantation of an artificial disc type in the L4/5 segment of the lumbar spine using a midline left-sided retroperitoneal approach. This approach included the mobilisation of the sympathetic trunk with incision and resection of the intervertebral disc. The perioperative and immediate postoperative periods were uneventful, but on the second postoperative day the patient complained of a progressive allodynia of the whole left leg in combination with weakness of the limb. Neurological examination did not reveal any radicular deficit or paresis. A sympathetic reaction following the mobilisation of the sympathetic trunk during the ventral preparation of the spine was suspected and investigated further. A diagnosis of CRPS I was proposed, and the patient was treated with analgesia, co-analgesics for pain alienation, and systemic corticosteroid therapy. A computed tomography-guided sympathetic block and lymphatic drainage were performed. Following conservative orthopaedic rehabilitation therapy, the degree of pain, allodynia, weakness, and swelling were reduced and the condition of the patient was ameliorated. The cost–benefit ratio of spinal arthroplasty is still controversial. The utility of this paper is to debate a possible cause of a painful complication, which can invalidate the results of a successful operation.

Keywords CRPS I · Artificial intervertebral disc surgery

S. M. Knoeller (✉)
Department of Orthopaedic and Trauma Surgery, University Hospital Freiburg, Freiburg, Germany
e-mail: stefan.knoeller@uniklinik-freiburg.de

S. M. Knoeller · B. Kleinmann · T. Wolter
Interdisciplinary Pain Centre, University Hospital Freiburg, Freiburg, Germany

M. Ehmer
Clinical-Ambulatory Pain Centre Freiburg, Freiburg, Germany

Introduction

Complex regional pain syndrome (CRPS) is a disease that is believed to be multifactorial involving the musculoskeletal and neurological systems which frequently occurs after a traumatic insult [4, 5, 18]. It was first described by Leriche in 1916 [21]. Different terms have been used for the pattern of CRPS, such as reflex sympathetic dystrophy syndrome (RSDS), Sudeck's atrophy, algodystrophia, and causalgia.

In 1993, the International Association for the Study of Pain (IASP) established the term that is accepted nowadays as "Complex Regional Pain Syndrome" (CRPS). Two main types are distinguished:

- CRPS type I (classical Sudeck's atrophy): symptoms of CRPS occur without any previous injury to the nerve.
- CPPS type II (causalgia): pain occurs as the result of an injury to a specific nerve, but is not essentially limited to the location of the injury.

Following the definition of IASP criteria, CRPS I is diagnosed by the following criteria:

- The presence of an initiating noxious event or a cause of immobilisation.
- Continuing pain, allodynia, or hyperalgesia, which is disproportionate to the causative event.
- Evidence of oedema, changes in skin blood flow, or abnormal sudomotor activity in the area of pain at some point in time.

- The diagnosis is excluded by the existence of any condition that would otherwise account for the degree of pain and dysfunction.

To the authors' knowledge, no case of CRPS I has been reported following the implantation of an artificial disc in the lumbar spine.

Case report

A 31-year-old Caucasian woman presented with chronic low back pain that had been present for 3 years. She had been treated previously with physiotherapy, analgesia, local anaesthetic injections, acupuncture and rest. She never became pain-free during this period, and even small movements led to persistent back pain. Plain X-rays showed osteochondrosis and sintering of the L4/L5 region (Fig. 1). A magnetic resonance imaging (MRI) scan revealed degeneration of the intervertebral disc at L4/5, and oedema in the endplates; no facet joint arthrosis was obvious (Fig. 2).

A diagnostic and therapeutic facet joint local anaesthetic block did not provide pain relief. A probatoric chest tube that was inserted for 1 week gave a degree of pain relief of about 80%. Discography of L4/5 evoked a typical "memory pain".

Therefore, there was an indication for the implantation of an artificial disc. The patient agreed to give consent after being fully informed about the procedure. The intervention was carried out under general anaesthesia. With the patient in a supine position, a 5-cm midline incision was made as

Fig. 2 An MRI image showing the degeneration of the L4/5 intervertebral disc and oedema of the endplates

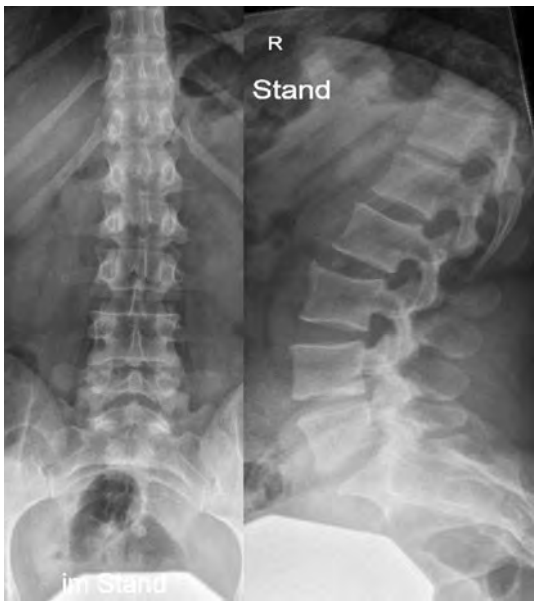

Fig. 1 Osteochondrosis and sintering L4/5

the initial part of a left-sided retroperitoneal approach to the L4/L5 intervertebral disc. After cautious mobilisation of the sympathetic trunk before incision of the longitudinal ligament, the intervertebral disc was resected and a medium-sized 10° Pro-Disc L was inserted. Intraoperative radiological scanning showed a perfect fit in both the anteroposterior and lateral views. The wound was closed and a 26-gauge Charr drain was inserted retroperitoneally. There was 30 ml of blood loss during the procedure, and the operation took 87 min in total. The immediate postoperative examination did not reveal any neurological deficit.

The patient was mobilised from the bed on the first postoperative day and this was uneventful; the retroperitoneal drainage was removed, and further X-rays that were taken in two planes with the patient in a standing position showed the correct position of the implant (Fig. 3).

On the second postoperative day, the patient complained of the onset of allodynia of the whole left leg in combination with weakness of the leg. The left leg appeared to be

Fig. 3 Postoperative control, Pro-Disc™ L4/5

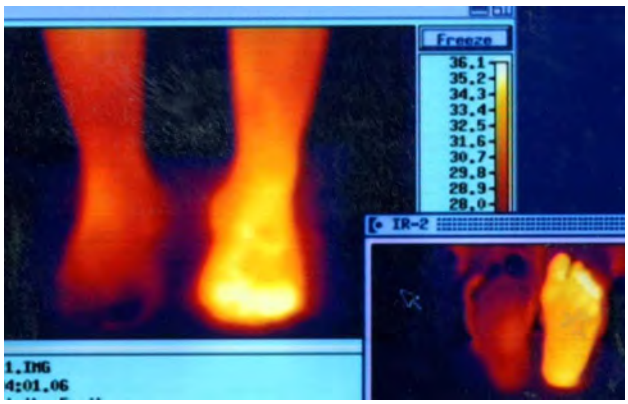

Fig. 4 Thermography: excessive hyperthermia of the left leg with accentuation on the foot

warmer than the right leg. An initial examination at that time did not reveal a neurological deficit, and a sympathetic reaction following the mobilisation of the sympathetic trunk during the procedure was suspected. Because of progressive allodynia in the left leg, further diagnostic investigations were initiated: an ultrasound examination and a computed tomography (CT) scan with contrast excluded a thrombosis affecting the left pelvis. Thermography (TG) (Fig. 4) showed excessive hyperthermia of the whole left leg, which particularly affected the foot; an ischaemia test was positive on the left side; a three-phase bone scan (Fig. 5a, b) showed increased nuclid storage in the left lower limb in all phases; and finally, magnetic resonance imaging (MRI) revealed slight oedema of the left lower extremity. Because of these findings and the allodynia, a diagnosis of CRPS I was made and immediate therapy was initiated. Treatment included analgesics using Oxycodone 1 × 10 mg and co-analgesics (Pregabalin 2 × 75 mg and Opipramol 2 × 50 mg) as well as systemic corticosteroid therapy with 1 × 80 mg 1,2-Dehydrocortisol for 5 days.

Because of the signs of a disturbance in the sympathetic nervous system, which included the increased temperature in the left leg, swelling and allodynia, lymphatic drainage and a CT-guided sympathetic block of the left sympathic trunk using 6 ml Bupivacaine 0.5% and 8 mg Dexamethasone was performed on two occasions, separated by an interval of 1 week (Fig. 6).

Following 2 weeks of hospital treatment, the pain, allodynia, weakness and swelling in the left leg had regressed; only a slight sensory disturbance of the left leg with pain on the dorsum of the foot with mild allodynia remained. A TG examination revealed persistent hyperthermia (Fig. 7). A conservative orthopaedic process of rehabilitation was successfully completed. One year after the surgery, the patient has a persistent discrete hyperaesthesia and allodynia of the left leg, which does not restrict her everyday life.

Discussion

Artificial disc surgery in the lumbar spine is an accepted method to treat refractory low back pain due to lumbar disc protrusion or degeneration. During surgery, mobilisation of the sympathetic trunk is necessary. Often, patients report hyperthermia of the extremity on the ipsilateral side of the retroperitoneal approach. This fact has been cited previously in the literature [44], where the patient reports that the contralateral leg is cooler than the ipsilateral leg, with respect to the side of the operative approach. Interestingly, patients usually notice and complain about the normal, unchanged leg as being “cool”, as opposed to the warm leg on the ipsilateral side of the surgery. Patients are informed of this risk prior to surgery as part of the informed consent procedure. To avoid this phenomenon, the sympathetic trunk should be carefully retracted and preserved as much as possible [39, 44].

Simmons has reported that this complication is temporary, and persists for between 3 and 4 months; he also states that it is rarely disabling. In the case presented here, the rapid, pronounced allodynia in combination with the pain and weakness without an obvious neurological deficit added a greater degree of complexity to this complication [10, 26, 27]. Diagnostic investigations for CRPS I in this patient showed unambiguous results and the chronological

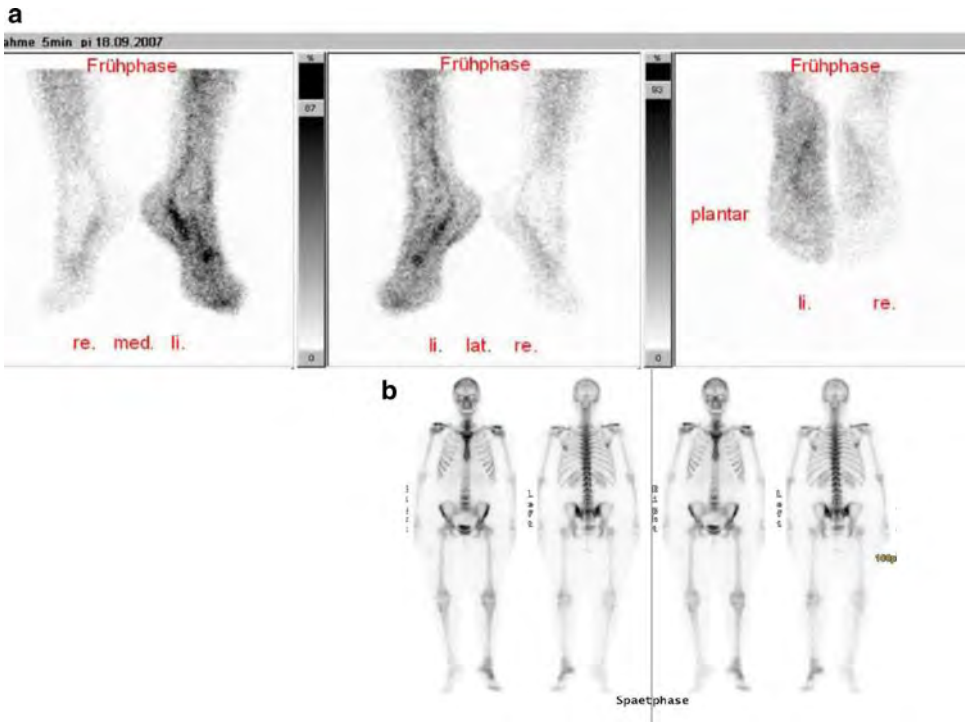

Fig. 5 Three-phase bone scan **a** early phase positive, **b** late phase positive

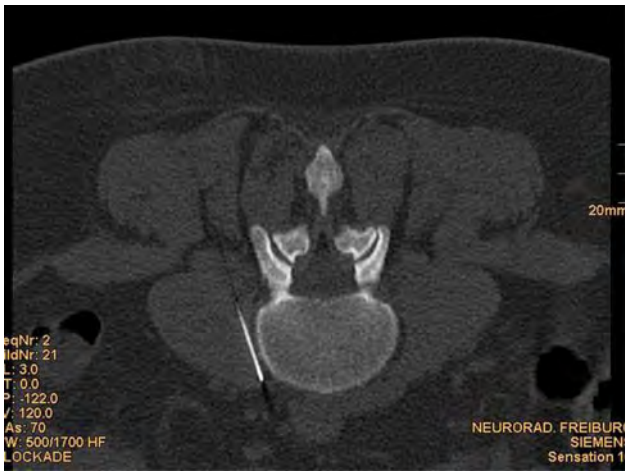

Fig. 6 CT guided blocation of the left sympathic trunk

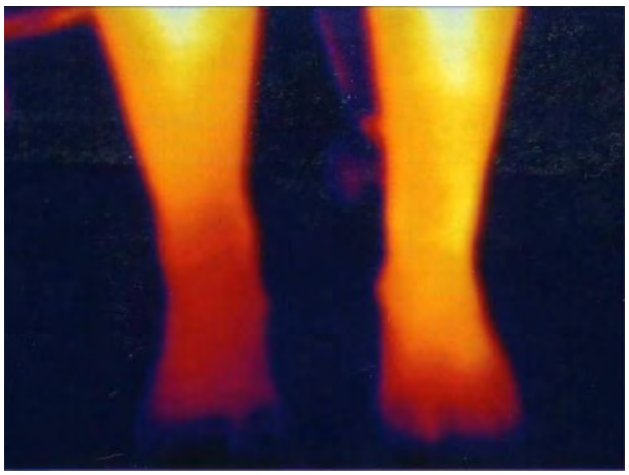

Fig. 7 Thermography control reveals considerable recurrent findings

coherence with surgery was an obvious aetiological feature of CRPS I in this case.

In summary, the diagnosis of CRPS I following artificial disc in the lumbar spine must be considered, and the decision was made to diagnose CRPS I instead of a specific sympathetic lesion due to the work of Veldman [41].

The delineation between a sympathetic reaction and CRPS is blurred. The controversy in the discrimination of a strong sympathetic reaction from CRPS has been discussed [32–34, 44]. Feldman et al. in 1993 [41] defined the diagnosis of CRPS if four of the following five criteria are found:

- inexplicable, diffuse pain
- change of the skin complexion
- diffuse oedema
- abnormal skin temperature
- limited activity of the affected extremity [2, 29, 30].

After a review of the literature, the diagnostic investigations for CRPS include laboratory exams [40], conventional X-rays of the affected extremity, TG, MRI [31], ischaemia tests [7] and bone scans [14, 19, 20, 25, 37]. Some authors advise body-plethysmography [36], laser colour duplex sonography and iontophoresis [38, 40].

Differential diagnoses of the patient in this case included thrombosis and subsequent blockage of the lymphatic drainage [1, 42, 43].

If the diagnosis of CRPS is made, prompt therapy is required as stasis and swelling of the affected limb or region leads to localised acidosis with propagation and perpetuation of CRPS [6]. Therapy includes systemic corticosteroids [3], pain medication following the World Health Organisation (WHO) III criteria [15, 28], in combination with co-analgesics for pain control (tricyclic antidepressants, anticonvulsants). Amelioration of the symptoms of CRPS has also been reported using calcitonin [17, 35] and calcium channel antagonists. Other therapies include physiotherapy, ergo therapy, manual lymphatic drainage, and sympathetic block of the stellatum in severe cases is sometimes required [8, 9].

In the literature, treatment options including spinal cord stimulation (SCS) [16], a guanethidine block [11, 12, 22], epidurals using morphine [23], ketamine comas and interventional therapies have been reported [24], although these were mostly reported in case reports and these therapies are not standard currently in the treatment of CRPS. Most reports indicate that the early initiation of a specific therapy leads to an earlier improvement of the disease [13, 18, 24, 34]. Functional capabilities may be affected due to CRPS if treatment is instituted in the later stages of the disease. The treatment described in this case report was specific for CRPS I, and an efficient improvement was gained with the CT-guided sympathetic block of the left sympathic trunk.

In summary, we report a case in which CRPS I presented following lumbar spine surgery via a ventral access, which must be distinguished from a sympathetic reaction due to the instrumental mobilisation of the sympathetic trunk. There is no further literature that has reported CRPS I following artificial disc surgery. Early diagnosis is very important as the early initiation of therapy in CRPS I can ameliorate the course of this disabling and potentially severe disease.

Conflict of interest None of the authors has any potential conflict of interest.

References

1. Albrecht PJ, Hines S, Eisenberg E, Pud D, Finlay DR, Connelly MK, Pare M, Davar G, Rice FL et al (2006) Pathologic alterations of cutaneous innervation and vasculature in affected limbs from patients with complex regional syndrome. Pain 120(3):244–266
2. Aronoff GM, Harden N, Stanton-Hicks M, Dorto AJ, Ensalada LH, Klimek EH, Mandel S, Wiliams JM (2002) American Academy of Disability Evaluating Physicians (AADEP) positions paper; complex regional pain syndrome 1 (RSD): impairment and disability issues. Pain Med 3(3):274–288
3. Bianchi C, Rossi S, Turi S, Brambilla A, Felisari G, Mascheri D (2006) Long-term functional outcome measures in corticosteroid-treated complex regional pain syndrome [In Process Citation]. Eura Medicophys 42(2):103–111
4. Birklein F (2005) Complex regional pain syndrome. J Neurol 252(2):131–138
5. Birklein F, Schmelz M, Schifter S, Weber M (2001) The important role of neuropeptides in complex regional pain syndrome. Neurology 57(12):2179–2184
6. Birklein F, Weber M, Ernst M, Riedl B, Neundorfer B, Handwerker HO (2000) Experimental tissue acidosis leads to increased pain in complex regional pain syndrome (CRPS). Pain 87(2):227–234
7. Blumberg H, Hoffmann U (1993) The so-called ischaemia test—a new diagnostic tool for reflex sympathetic dystrophy. Schmerz 7(3):178–181
8. Bonelli S, Conoscente F, Movillia PG, Restelli L, Francucci B, Grossi E (1983) Regional intravenous guanethidine vs stellate ganglion block in reflex sympathetic dystrophies: a randomized trial. Pain 16(3):297–307
9. Dunningham TH (1980) The treatment of Sudeck′s atrophy in the upper limb by sympathetic blockade. Injury 12(2):139–144
10. Gradl G, Schurmann M (2005) Sympathetic dysfunction as a temporary phenomenon in acute posttraumatic CRPS 1. Clin Auton Res 15(1):29–34
11. Hannigton-Kiff JG (1974) Intravenous regional sympathetic block with guanethidine. Lancet 1(7865):1019–1020
12. Hannington-Kiff JG (1977) Relief of Sudeck‘s atrophy by regional intravenous guanethidine. Lancet 1(8022):1132–1133
13. Harden RN, Swan M et al (2006) Treatment of complex regional pain syndrome: functional restoration. Clin J Pain 22(5):420–424
14. Intenzo C, Kim S, Millin J, Park C (1989) Scintigraphic patterns of the reflex sympathetic dystrophy syndrome of the lower extremities. Clin Nucl Med 14(9):657–661
15. Kalita J, Vajpayee A, Misra UK (2006) Comparison of prednisolone with piroxicam in complex regional pain syndrome following stroke: a randomized controlled trail. QJM 99(2):89–95
16. Kemler MA, Barendse GA, van Kleef M, de Vet HC, Rijks CP, Furnee CA, van den Wildenberg FA (2000) Spinal cord stimulation in patients with chronic reflex sympathetic dystrophy. N Engl J Med 343(9):618–624
17. Kissling RO, Bloesch AC, Sager M, Dambacher MA, Schreiber A (1991) Prevention de la recidive d′une maladie de Sudeck par la calcitonine. Rev Chir Orthop Reparatrice Appar Mot 77(8):562–567
18. Kock FX, Borisch N, Koester B, Grifka J (2003) Complex regional pain syndrome type 1 (CRPS 1). Pathophysiology diagnostics and therapy. Orthopade 32(5):418–431
19. Kozin F, Ryan LM, Carrera GF, Soin JS, Wortmann RL (1981) The reflex sympathetic dystrophy syndrome (RSDS) III. Scintigraphic studies, further evidence for the therapeutic efficacy corticosteroids, and proposed diagnostic criteria. Am J Med 70(1):23–30
20. Kozin F, Soin JS, Ryan LM, Carrera GF, Wortmann RL (1981) Bone scintigraphy in the reflex sympathetic dystrophy syndrome. Radiology 138(2):437–443
21. Leriche R (1916) De la causalgie, envisage comme une nevrite du sympathique et de son traitement par la denudation et l‘excision des plexus nerveux periartiels. Presse Med 24:178–180
22. Livingstone JA, Atkins RM (2002) Intravenous regional guanethidine blockade in the treatment of post-traumatic complex regional syndrome type 1 (algodystrophy) of the hand. J Bone Joint Surg Br 84(3):380–386
23. Moufawad S, Malak O, Mekhail NA (2002) Epidural infusion of opiates and local anesthetics for Complex Regional Pain Syndrome. Pain Pract 2(2):81–86

24. Nelson DV, Stacey BR (2006) Interventional therapies in the management of complex regional pain syndrome. Clin J Pain 22(5):438–442
25. Pankaj A, Kotwal PP, Mittal R, Deepak KK, Bal CS (2006) Diagnosis of post-traumatic complex regional pain syndrome of the hand: current role of sympathetic skin response an three-phase bone scintigraphy. J Orthop Surg 14(3):284–290
26. Rommel O, Gehling M, Dertwinkel R, Witscher K, Zenz M, Malin JP, Janig W (1999) Hemisensory impairment in patients with complex regional pain syndrome. Pain 80(1–2):95–101
27. Rommel O, Malin JP, Zenz M, Janig W (2001) Quantitative sensory testing, neurophysiological and psychological examination in patients with complex regional pain syndrome and hemisensory deficits. Pain 93(3):279–293
28. Rowbotham MC (2006) Pharmacologic management of complex regional pain syndrome. Clin J Pain 22(5):425–429
29. Schasfoort FC, Bussmann JB, Krijnen HJ, Stam HJ (2006) Upper limb activity over time in complex regional pain syndrome type I as objectively measured with an upper limb-activity monitor: an explorative multiple case study. Eur J Pain 10(1):31–39
30. Schasfoort FC, Bussmann JB, Stam HJ (2004) Impairment and activity limitations in subjects with chronic upper-limb complex regional pain syndrome type 1. Arch Phys Med Rehabil 85(4):557–577
31. Schimmerl S, Schurawitzki H, Imhof H, Canigiani G, Kramer J, Fialka V (1991) Morbus Sudeck–MRT als neues diagnostisches Verfahren. Rofo Fortscher Geb Rontgenstr Neunen Bildgeb Verfahr 154(6):601–604
32. Schinkel C, Gaertner A, Zaspel J, Zedler S, Faist E, Schuermann M (2006) Inflammatory mediators are altered in the acute phase of posttraumatic complex regional pain syndrome. Clin J Pain 22(3):235–239
33. Schürmann M, Gradl G, Zaspel J, Kayser M, Löhr P, Andress HJ (2000) Peripheral sympathetic functions as a predictor of complex regional pain syndrome type 1 (CRPS) in patients with radial fracture. Auton Neurosci 86(1–2):127–134
34. Schürmann M, Gradl G, Rommel O (2007) Early diagnosis in post-traumatic complex regional pain syndrome. Orthopedics 30(6):450–456
35. Schürmann M, Vogel T, Gärtner A, Andress H-J, Gradl G (2001) Erfahrungen mit der Kalzitonin-Behandlung bei Patienten mit Complex Regional Pain Syndrome Type 1 (CRPS 1 – M. Sudeck). Z Orthop 139:452–457
36. Schürmann M, Zaspel J, Gradl G, Wipfel A, Christ F (2001) Assessment of the peripheral microcirculation using computer-assisted congestion plethysmography in post-traumatic Complex Regional Pain Syndrome type 1. J Vasc Res 38(5):453–461
37. Shehab D, Elgazzar A, Collier BD, Naddaf S, Al Jarallah K, Omar A, Al-Murtairy M (2006) Impacht de three-phase bone scintigraphy on the diagnosis and treatment of complex regional pain syndrome type 1 or reflex sympathetic dystrophy. Med Princ Pract 15(1):46–51
38. Shenker NG, Mapp PI, Harris ND, Blake DR (2005) Assessment of endothelial functions in complex regional pain syndrome type 1 using iontophoresis and laser Doppler imaging. Rheumatology 44(2):264–265
39. Simmons EH, Trammell TR (1983) Operative management of adult scoliosis. In: Evarts MC (ed) Scoliosis of musculoskeletal system. Churchill Livingstone, New York
40. Tan EC, Oyen WJ, Goris RJ (2005) Leukocytes in complex regional pain syndrome type. 1 Inflammation 29(4–6):182–186
41. Veldman PH, Reynen HM, Arntz IE, Goris RJ (1983) Signs and symptoms of reflex sympathetic dystrophy: prospective study of 829 patients. Lancet 342(8878):1012–1016
42. Wasner G, Heckmann K, Maier C, Baron R (1999) Vascular abnormalities in acute reflex sympathetic dystrophy (CRPS): complete inhibition of sympathetic nerve activity with recovery. Arch Neurol 56(5):613–620
43. Wasner G, Schattschneider J, Baron R (2002) Skin temperature side differences—a diagnostic tool for CRPS? Pain 98(1–2):19–26
44. Zdeblick T (1999) Anterior approaches to the spine. Quality Medical Publishing, St Louis, p 166