



Complex regional pain syndrome as a result of total knee arthroplasty: A case report and review of literature

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ABSTRACT

Total knee arthroplasty (TKA) is an effective treatment for patients with end-stage symptomatic knee osteoarthritis. As the aging population grows, the demand for the procedure is projected to increase. While highly successful, TKA has associated risks and complications. Complex regional pain syndrome is one uncommon but debilitating complication that can negatively impact patient satisfaction and quality of life. We present a case of complex regional pain syndrome in the operated leg that resulted in significant functional deficits. Key findings of this case include significant and disproportionate pain in the joint, altered cutaneous sensation around the joint, and decreased range of motion in flexion in the absence of any mechanical issues with the TKA. Because of the debilitating nature of this condition, patients must be fully informed of and realize the risks associated with undergoing a widely appreciated procedure such as TKA.

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1. Introduction

Knee osteoarthritis causes significant declines in quality of life due to pain and loss of function. Total knee arthroplasty (TKA) is a highly successful treatment for osteoarthritis [1]. Concomitant with increasing life expectancies and an aging population, the demand for TKA in the U.S. is projected to increase by 143% between 2012 and 2050; the number of TKA cases is expected to reach 1.5 million per year by 2050 [2]. Although TKA is regarded as the gold-standard treatment for osteoarthritis, the procedure may fall short of patients' high expectations in terms of total pain relief [3]. Postoperatively, residual pain is a strong predictor of patient dissatisfaction with the procedure [3].

An uncommon debilitating side-effect of TKA is complex regional pain syndrome (CRPS). CRPS is characterized by a chronic burning sensation at or distal to a site of physical trauma which exceeds the expected severity and duration of the initial physical trauma, with little or no improvement over time. It is commonly associated with discoloration, temperature and trophic changes, and edema at or distal to the trauma site [4]. The types of trauma that precede CRPS are typically either fractures or surgical procedures [5]. Although improvements in flexion are the expected result of TKA, the improvements are diminished in CRPS patients compared to patients without CRPS [6]. While

an uncommon complication of TKA, CRPS should be included in the differential diagnoses of patients who recover slowly with greater than expected pain when other causes have been excluded. Early treatment may prevent undesirable outcomes and improve patient satisfaction with the procedure [6].

This report will discuss the case of a patient who underwent TKA to treat end-stage knee osteoarthritis and was later diagnosed with CRPS.

2. Case information

A 69-year-old woman presented to her general practitioner with pre-existing pain in her right knee that worsened following a twisting injury. No ligamentous injury was detected and her range of motion was 10° to 100° of flexion. Analgesia and physiotherapy were ineffective, and she was referred to orthopaedics. End-stage knee osteoarthritis was diagnosed.

The patient underwent right TKA one month after diagnosis, experienced no perioperative or immediate postoperative complications, and was discharged after four days. She was prescribed discharge medications as per the local protocol, including regular paracetamol and dihydrocodeine as required for management. At one-month telephone follow-up there had been no postoperative complications. The patient continued exercises as directed for rehabilitation but still experienced residual swelling.

Two months postoperatively, the patient had pain and discomfort in the operated knee, and this was associated with altered sensations in the leg along with hypersensitivity to touch (hyperalgesia). The patient

Abbreviations: TKA, Total knee arthroplasty; CRPS, Complex regional pain syndrome.

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also mentioned that she could not tolerate even slight touch or a bed sheet over that knee (allodynia). Blood tests as well as repeat imaging produced no evidence of infection or component loosening. These features are typical of CRPS, for which she was prescribed gabapentin 300 mg once per day. As the patient did not respond to the initial dose, gabapentin was increased to 300 mg three times per day, which did somewhat reduce the pain. Nonetheless, the patient continued to note a throbbing, burning sensation in the posterior aspect of her knee and hyperalgesia and paresthesia on light touch over the lateral aspect. Range of motion was restricted by pain to 10° to 70° of flexion. The patient was referred to physiotherapy to help restore knee movement, analgesia was continued, and a pain consultant was contacted to address her CRPS. The patient continued to struggle with CRPS, despite slight improvements in pain and in range of motion, which was from 10° to 90° of flexion with discomfort at the last follow-up, five months postoperatively.

3. Discussion

CRPS is an uncommon but debilitating condition. There are two categories of CRPS: CRPS-I, also known as reflex sympathetic dystrophy (RSD), refers to cases where peripheral nerve injury is absent; and CRPS-II, also known as causalgia, which refers to cases where nerve damage is present [7]. The estimated incidence of CRPS in the U.S. is 20,000–80,000 per year with a greater proportion of women affected than men [4]. One study found that elective surgery was the primary cause of 12% of the 1043 CRPS cases analyzed [8]. The relatively low incidence rate associated with elective surgery may lead to missed diagnoses of CRPS and diminished patient satisfaction [6]. The time of onset of CRPS symptoms typically ranges from immediately to several weeks postoperatively, but diagnosis is often delayed [9]. The patient in this case report appears to have experienced characteristic symptoms over this time range but did not receive a diagnosis of CRPS until over 3 months after her TKA.

The pathophysiology of CRPS is unclear but several mechanisms have been proposed: neuroplastic changes, inflammatory processes, and vasomotor functional deficits. Changes in peripheral nociceptors and decreased cortical regulation may lead to a disproportionate response to little or no pain stimulus [10]. Inflammation caused by the initial trauma results in the release of neuropeptides that may also cause a long-term increase in pain sensitivity and edema [10]. Vasomotor deficits caused by an accumulation of catecholamines result in characteristic temperature changes and may increase nociceptive activation and sensitization [10]. While these proposed mechanisms provide possible explanations for the manifestation of CRPS, the lack of a conclusive pathophysiological mechanism contributes to the difficulty of effectively managing the condition.

Interdisciplinary management is key for CRPS. Patient education, pain relief, physical and occupational rehabilitation, and psychological intervention are central in treating CRPS [11]. Physiotherapy is used to restore function in conjunction with medications for the associated pain [12]. In this case, the patient was referred to physiotherapy and prescribed up to 900 mg gabapentin per day to address her CRPS. The patient was unable to tolerate physiotherapy due to pain. Gabapentin, which has been found to be moderately effective in providing pain relief in the earlier stages of CRPS, was prescribed with the aim of decreasing neuronal excitability, as CRPS is considered a neuropathic pain condition [13]. Unfortunately, the patient in this case found gabapentin to be of limited effectiveness. Lumbar sympathetic nerve blocks and sympathectomies are available as additional treatment options, though their long-term effectiveness is inconclusive and complications are common [14].

People with CRPS of the knee typically experience limitations in their ability to perform moderately physically demanding tasks, due to pain [15]. The patient in this case noted having to adjust basic tasks to avoid pain. On physical examination, pain restricted knee flexion. An

interesting aspect of this case is that the patient's range of motion in flexion was worse after TKA than before the procedure due to CRPS, whereas TKA typically results in improved range of motion in flexion, even in patients later diagnosed with CRPS [6]. At the time of writing the patient was awaiting a consultation with a pain specialist and had the option of undergoing manipulation under anesthesia to restore range of motion in her operated knee if other measures were ineffective.

CRPS places a significant burden on patients postoperatively and negatively impacts overall satisfaction with procedures such as TKA. One study [16] found that only one-third of its participants diagnosed with CRPS experienced full resolution of their condition. Further, the literature notes that 19% of patients who undergo TKA are indifferent to or dissatisfied with the outcome [17], which may be due to unrealistic expectations in terms of pain relief and restoration of function [3].

The patient in this case emphasized her regret of undergoing TKA, as she noted that her quality of life was better before the procedure. This case highlights the importance of informing patients that, while TKA is generally a very successful intervention for osteoarthritis, there is a risk of no improvement or worsening of pain and overall functional deficits after the procedure. Providing further clarity to the patient regarding possible outcomes of TKA such as CRPS may help to ameliorate misalignments between patient expectations and the end result of surgical interventions.

Contributors

J Matthew Royeca wrote the draft, conducted the literature search, and interviewed the patient.

All other co-authors contributed equally, including to the editing of the draft, and saw and approved the final version.

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Patient consent

Informed consent was obtained from the patient.

Provenance and peer review

This case report was peer reviewed.

Declaration of Competing Interest

The authors declare that they have no conflict of interest regarding the publication of this case report.

References

- [1] Total knee replacement: an evidence-based analysis, *Ont. Health Technol. Assess. Ser.* 5 (9) (2005) 1–51.
- [2] M.C.S. Inacio, et al., Projected increase in total knee arthroplasty in the United States – an alternative projection model, *Osteoarthr. Cartil.* 25 (11) (2017) 1797–1803, <https://doi.org/10.1016/j.joca.2017.07.022>.
- [3] P.N. Baker, et al., The role of pain and function in determining patient satisfaction after total knee replacement. Data from the National Joint Registry for England and Wales, *J. Bone Joint Surg. (Br.)* 89 (7) (2007) 893–900, <https://doi.org/10.1302/0301-620X.89B7.19091>.
- [4] J. Marinus, et al., Clinical features and pathophysiology of complex regional pain syndrome, *Lancet Neurol.* 10 (7) (2011) 637–648, [https://doi.org/10.1016/S1474-4422\(11\)70106-5](https://doi.org/10.1016/S1474-4422(11)70106-5).
- [5] K.D. Bharwani, M. Dirckx, F.J. Huygen, Complex regional pain syndrome: diagnosis and treatment, *BJA Educ.* 17 (8) (2017) 262–268, <https://doi.org/10.1093/bjaed/mlx007>.
- [6] A.W. Burns, et al., Complex regional pain syndrome complicating total knee arthroplasty, *J. Orthop. Surg. (Hong Kong)* 14 (3) (2006) 280–283, <https://doi.org/10.1177/230949900601400309>.

- [7] R.N. Harden, et al., Proposed new diagnostic criteria for complex regional pain syndrome, *Pain Med.* 8 (4) (2007) 326–331, <https://doi.org/10.1111/j.1526-4637.2006.00169.x>.
- [8] S. Ott, C. Maihofner, Signs and symptoms in 1,043 patients with complex regional pain syndrome, *J. Pain* 19 (6) (2018) 599–611, <https://doi.org/10.1016/j.jpain.2018.01.004>.
- [9] Z. Li, et al., Complex regional pain syndrome after hand surgery, *Hand Clin.* 26 (2) (2010) 281–289, <https://doi.org/10.1016/j.hcl.2009.11.001>.
- [10] S. Bruehl, An update on the pathophysiology of complex regional pain syndrome, *Anesthesiology* 113 (3) (2010) 713–725, <https://doi.org/10.1097/ALN.0b013e3181e3db38>.
- [11] L. Turner-Stokes, A. Goebel, Complex regional pain syndrome in adults: concise guidance, *Clin. Med. (Lond.)* 11 (6) (2011) 596–600, <https://doi.org/10.7861/clinmedicine.11-6-596>.
- [12] R.S. Perez, et al., Evidence based guidelines for complex regional pain syndrome type 1, *BMC Neurol.* 10 (2010) 20, <https://doi.org/10.1186/1471-2377-10-20>.
- [13] A.K. Tan, et al., The effect of gabapentin in earlier stage of reflex sympathetic dystrophy, *Clin. Rheumatol.* 26 (4) (2007) 561–565, <https://doi.org/10.1007/s10067-006-0350-y>.
- [14] S. Straube, et al., Cervico-thoracic or lumbar sympathectomy for neuropathic pain and complex regional pain syndrome, *Cochrane Database Syst. Rev.* (9) (2013) Cd002918, <https://doi.org/10.1002/14651858.CD002918.pub3>.
- [15] C.M. van Bussel, D.L. Stronks, F. Huygen, Clinical course and impact of complex regional pain syndrome confined to the knee, *Pain Med.* (2019) <https://doi.org/10.1093/pm/pnz002>.
- [16] M. de Mos, et al., Outcome of the complex regional pain syndrome, *Clin. J. Pain* 25 (7) (2009) 590–597, <https://doi.org/10.1097/AJP.0b013e3181a11623>.
- [17] R.B. Bourne, et al., Patient satisfaction after total knee arthroplasty: who is satisfied and who is not? *Clin. Orthop. Relat. Res.* 468 (1) (2010) 57–63, <https://doi.org/10.1007/s11999-009-1119-9>.