



Clinical pain research

Complex regional pain syndrome following viper-bite

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ABSTRACT

Background: Complex regional pain syndrome (CRPS) may occur following fractures, surgery or different trauma. Development of CRPS following snake-bite has only been published in three reports (from Turkey, Nepal and Korea), although snake bites occur frequently world-wide. There has been no report from Western Europe. *Vipera Berus* is a common snake in European countries and the only venomous snake in Norway. We here describe the development of CRPS in a young woman as a consequence of a viper bite (*Vipera Berus*) in the right arm.

Methods: We performed a clinical investigation (inspection, measurement of skin temperatures, sensory and motor evaluation) of the patient six months following the viper-bite, measurement of thermal thresholds (quantitative sensory testing, QST), measurement of resting sweat output (RSO) and quantitative sudomotor axon reflex (QSART) from both arms.

Results: The patient fulfilled the Budapest criteria for a CRPS-condition, with continuous pain and symptoms and findings of autonomic dysfunction. In addition, we found elevated thresholds of warmth and cold, evidence of an affection of afferent A-delta and C-fibres as well as an affection of the efferent sympathetic sudomotor C-fibres by QSART. An increased RSO-volume was in inverse relationship to the decreased QSART result.

Conclusion and Implications: It is important to be aware of viper-bite as a possible eliciting event for CRPS for early diagnosis and treatment of a patient. As long-lasting pain and oedema are known complications, it is probable that CRPS after viper-bites previously may have been underdiagnosed. As many patients are unaware of being bit, viper bite should be considered in cases of unexplained sudden pain and swelling of a limb.

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1. Introduction

Complex regional pain syndrome (CRPS) is a serious and disabling chronic pain condition, usually occurring in a limb. The clinical picture is dominated by pain and symptoms of autonomic dysfunction, but may also involve motor disability. Complex regional pain syndrome (CRPS) has a reported incidence up to 26 per 100,000 person years [1]. CRPS may develop as a consequence of an injury, most frequently following fractures [1,2], but may also occur without any known eliciting event [3]. Despite the high number of snake-bites worldwide, CRPS are described only in three case-reports (from Turkey, Nepal and Korea) [4–6]. CRPS after snake

bites in Western Europe has to our knowledge not previously been reported.

2. Case story

The patient is a 22 year old woman, previously healthy and with no family history of chronic pain syndromes. She was prior to the incident working full time as a cook. On October the 7th 2014, she discovered a discolouring of the skin in the right arm, above the elbow up to the shoulder. Simultaneously, she developed a deep aching pain in the same area, rapidly expanding to involve the whole right arm. She consulted her private doctor (Dr. Skulberg) who noticed two bites of viper in the elbow region. She recalled having thrown a bag of garbage in a bin, situated on the edge of a wood two days prior to the appearance of her symptoms. The red discolouring of the skin gradually disappeared within a week while the pain continued. She was treated with cortisone. In addition to the spontaneously ongoing aching pain, she also suffered from paroxysmal pain in the whole right arm, of short duration

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Fig. 1. Picture of discolouring of skin a few days after viper bite.

3. Methods

We performed a clinical investigation, quantitative sensory testing (QST; measurement of thermal detection thresholds), measurement of spontaneous sweating (resting sweat output (RSO)) and quantitative sudomotor axon reflex (QSART).

3.1. Clinical investigation

We looked for possible oedema, discoloration of skin, glossy or sweaty skin, signs of motor dysfunction (involuntary movement, atrophy), measured skin temperature at three different sites right arm (thenar-eminence, at the level of the elbow and shoulder (with the use of Somedic's Tempett (Hörby, Sweden)) and performed a neurological examination with an emphasis of analysis of motor and sensory function, including testing for allodynia to light touch and hyperalgesia to pin-prick and blunt pressure.

4. Quantitative sensory testing (thermal detection thresholds)

Threshold temperatures for the sensation of warmth, cold, heat pain and cold pain were determined using a computerized Thermotest (SENSElab, Somedic A/B, Hörby, Sweden) with a thermode size of 5 cm × 2.5 cm. Warmth detection threshold (WD), cold detection threshold (CD), heat pain detection threshold (HP) and cold pain detection threshold (CP) were determined from a baseline temperature of 32° C with a 1 °C/s rate of change. The patient was instructed to push a signal-button when the relevant sensation was perceived. When this happened or if cut-off temperature (50° C and 10° C) was obtained, the temperature returned to baseline. WD, CD, HP, CP were determined from the thenar eminence, the region of the elbow and the upper arm. For calculations of thresholds, an average of five recordings for WD and CD and the average of three recordings for HP and CP were used. The values were compared with the healthy left arm.

5. QSART

Evoked sweat response at rest was measured by the Quantitative sudomotor axon reflex tester (WR Medical Electronics Co., Stillwater, USA). It turned impossible to perform the investigation from the palm of the hand as intended, and we therefore had to investigate the dorsum of the hand.

To evoke axon reflex sweating, iontophoresis (2 mA for 5 min) of 10% acetylcholine (ACh) was started after a stable baseline sweating level had been obtained. A commercially available device (Iontophor II, Life-Tech Inc, Stafford, TX, USA) was used for this purpose. Axon reflex sweat output during the 5 min of iontophoresis and additional 10 min was recorded via a sweat capsule with an area of 0.767 cm² attached to the skin of the back of both hands.

5.1. Resting sweat output (RSO)

We measured spontaneous sweat output with the same equipment as for QSART ((see above) but with no iontophoresis of Ach) from the thenar eminence of both hands.

6. Results

6.1. Clinical investigation

There were sidelike and normal findings upon inspection, no oedema, no trophic or colour changes of skin, no obvious increased sweating in the right hand, no atrophy or other motor

(seconds) and with a stinging character. She had no allodynia to light touch or hyperalgesia to pin-prick/blunt pressure in this early stage.

She was during the two following months on 100% sick-leave and the pain gradually diminished but did not disappear. The pain worsened as she returned to work, first 50% in 1½ month and 100% from January 2015.

From December 2014 she developed an intermittent oedema in the fingers and the radial part of the lower right arm, appearing with duration of one day each 1–3 weeks, from January 2015 with a simultaneous blue discolouring of the skin, from the back of the hand and lower arm up to the level of the elbow. She also noticed that her hand often was cold, and if not cold it was moist with an excessive abnormal sweating, the sweat drops dripping from her hand. She had neither noticed additional autonomic dysfunctions nor any motor problems. Prior to the investigation at OUS-Rikshospitalet, EMG/neurography had proven normal. The patient consents to the publication of her experience (Fig. 1).

2.1. Symptoms at time of investigation

At the day of investigation (April 7, 2015) at the Section of Clinical Neurophysiology, Oslo University Hospital, she complained about a spontaneous and constantly ongoing pain of a mainly aching character, but with some soreness, deep in the whole right arm with an intensity of minimum 6 and maximum 10 (on a numeric scale ranging from 0 to 10, 0 being no pain and 10 worst imaginable pain). The intensity of the pain could increase spontaneously, but did always increase following use of the arm, which rendered her work impossible (she is right-handed). She had experienced an abnormal pain by pressure of the arm and also following needle-stick (taking of blood-sample).

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