

Clinical case

Double crush syndrome of the median nerve revealing a primary non-Hodgkin's lymphoma of the flexor digitorum superficialis muscle

Syndrome de double compression du nerf médian révélant une localisation primaire d'un lymphome non hodgkinien au muscle flexor digitorum superficialis

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Abstract

Extranodal manifestations of lymphoma are well described in the literature and occur in 20 to 30% of patients. Skeletal muscle involvement is rare. We describe the case of a patient with non-Hodgkin's lymphoma in a forearm muscle. At the age of 86, the featured patient started experiencing continuous, progressive and high intensity pain that was more frequent at night and localized in the right dominant hand. It was associated with paresthesia and hypoesthesia, primarily in the thumb, index finger and middle finger. Clinical examination and electrodiagnosis led to the diagnosis of carpal tunnel syndrome. The patient underwent carpal tunnel release at a private hand center. The progression was unfavorable. Additional clinical examination and electrodiagnosis showed compression of the anterior interosseous nerve (double crush syndrome). The patient was referred to our university hand center for further management. Magnetic resonance imaging showed a large mass of about 20 cm occupying the entire anterior compartment of the forearm and enclosing the median nerve. Biopsies were performed and revealed a diffuse large B-cell primary non-Hodgkin's lymphoma. The patient underwent chemotherapy and radiotherapy. Six months later, the patient was in complete remission. Muscular involvement during lymphoma is rare. Biopsy is mandatory; needless radical surgery can be avoided because lymphoma is primarily a non-surgical disease. The key points of the treatment process are reviewed.

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Keywords: Double crush syndrome; Median nerve; Primary lymphoma; Non-Hodgkin's lymphoma

Résumé

Les manifestations extralymphonodales du lymphome sont bien décrites dans la littérature et se produisent chez 20 à 30 % des patients. L'implication des muscles squelettiques est rare. Nous décrivons le cas d'un patient ayant un lymphome non hodgkinien avec localisation musculaire à l'avant-bras. À l'âge de 86 ans, le patient avait commencé à ressentir des douleurs continues, progressives et de haute intensité, plus fréquemment la nuit et localisées à la main droite (dominante). Ces douleurs étaient associées à des paresthésies et une hypoesthésie localisées principalement au pouce, à l'index et au majeur. L'examen clinique et électrique avait mis en évidence un syndrome du canal carpien. Le patient avait bénéficié d'une libération du nerf médian au canal carpien dans un centre privé de chirurgie de la main. L'évolution avait été défavorable. L'examen clinique et l'examen électrophysiologique avaient montré une compression du nerf interosseux antérieur. Le patient a été pris en charge dans notre centre universitaire de chirurgie de la main. L'imagerie par résonance magnétique a montré une volumineuse masse de 20 cm de hauteur occupant l'ensemble du compartiment antérieur de l'avant-bras et entourant le nerf médian. Les biopsies effectuées ont révélé un lymphome non hodgkinien diffus à grandes cellules B. Le patient a reçu une chimiothérapie et une radiothérapie. Six mois plus tard, le patient était en rémission.

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complète. La discussion revient sur les points clés de la prise en charge. La participation musculaire dans le cadre du lymphome est rare. La biopsie est obligatoire et permet d'éviter une chirurgie radicale inutile.

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Mots clés : Syndrome de double compression nerveuse ; Nerf médian ; Lymphome primaire ; Lymphome non hodgkinien

1. Introduction

Extranodal manifestations of lymphoma are well described in the literature and occur in 20 to 30% of patients, with the incidence increasing in recent decades [1]. Skeletal muscle involvement has been reported only rarely. In the literature, these cases represent only 0.1 to 2.0% of the extranodal lymphomas [2–5]. The mechanism for muscle involvement in malignant lymphoma is mostly secondary to direct extension from adjacent lymph nodes and bone or by dissemination; however, primary muscular involvement has been described as well [6]. The most frequently affected muscles are the gluteal and pelvic muscles [3].

We present the clinical history and magnetic resonance imaging findings in a case of double crush syndrome of the median nerve associated with extranodal non-Hodgkin's lymphoma (NHL) in the forearm region involving primarily the muscles of the anterior compartment.

2. Case report

An 86-year-old woman started experiencing continuous, progressive and high intensity pain and dysesthesia that was more frequent at night and localized in the right dominant hand. It was associated with paresthesia and hypoesthesia, primarily of the thumb, index finger and middle finger. Clinical examination and electrodiagnosis led to diagnosis of carpal tunnel syndrome (CTS). The patient underwent surgical carpal tunnel release in October 2011 at a private hand center. Despite physiotherapy, her progression was unfavorable: a few weeks after surgery, she noticed a reduction in the numbness but the onset of symptoms that were not present earlier, such as inability to flex the thumb, index and middle finger. The pain was constant and seemed to involve the entire forearm. Additional clinical examination and electrodiagnosis revealed complete denervation of the flexor pollicis longus (FPL), the flexor digitorum profundus (FDP) of the index and middle fingers including the pronator quadratus (PQ), suggestive of anterior interosseous (AIN) nerve syndrome.

The patient was referred to our university hand center for further management in May 2012. Generally, she had no fever or sweating. She had a sharp decrease in appetite and had lost 25 kg in two months from 115 to 90 kg (1.55 m height). On physical examination, she was unable to actively flex the interphalangeal joint of the right thumb and distal interphalangeal joints of the right index and middle fingers. The forearm showed no obvious signs of edema. Pain was spontaneous and constant but increased on palpation. There was a positive Tinel's sign and reduced grip strength (72% of the contralateral side). Preoperative complete blood count and chest X-ray were

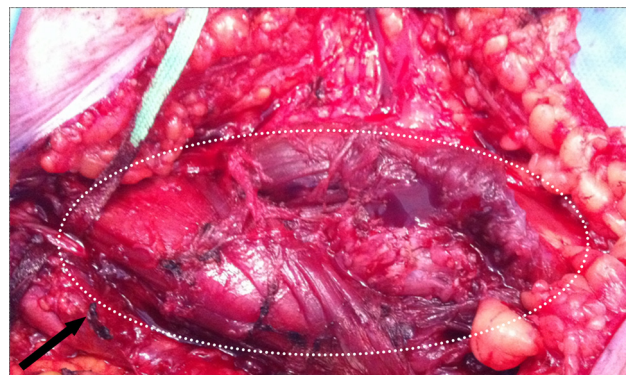


Fig. 1. Intraoperative view of the tissue mass invading muscle; proximal part of median nerve (black arrow).

unremarkable. On June 2012, during surgical exploration, the flexor digitorum superficialis muscle was tight, elevated and invaded by a suspicious fibrous mass (Fig. 1), which was found to encompass the median nerve and its branch, the AIN. Biopsies revealed a diffuse large B-cell primary non-Hodgkin's lymphoma of the flexor digitorum superficialis muscle (Fig. 2).

Magnetic resonance imaging (MRI) showed a large tumor mass (Fig. 3) of about 20 cm occupying the entire anterior compartment of the forearm and enclosing the median nerve. The tumor's boundaries were just above the carpal tunnel distally and at the elbow without joint invasion proximally. A posterior extension of the tumor came into contact with the antebrachial interosseous membrane. Postoperative staging investigations included echocardiography, bone marrow

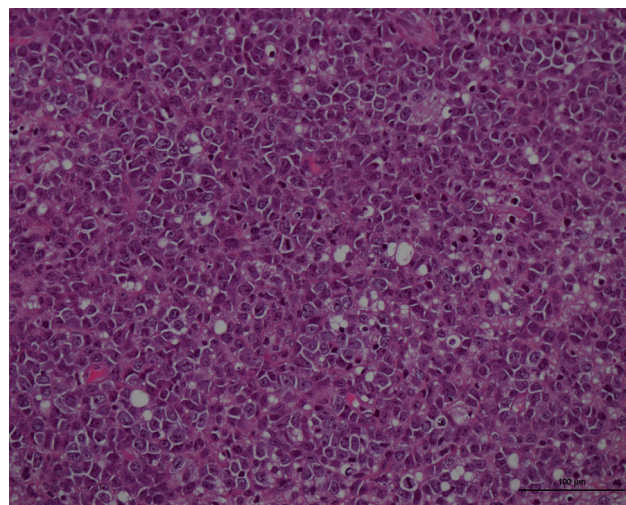


Fig. 2. High power view of the tumor proliferation consisting of numerous large primitive cells having the appearance of a star-filled sky. These cells are not cohesive.

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