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Carpal tunnel syndrome due to hibernoma of the wrist: Case report

Syndrome du canal carpien secondaire à un hibernome du poignet : cas clinique

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ABSTRACT

Carpal tunnel syndrome (CTS) is the most frequent among entrapment neuropathies. The usual etiology is idiopathic, but can be caused by space-occupying lesions, a relatively rare condition. This condition becomes exceptional when the space-occupying lesion corresponds to a rare tumor whose location is atypical. The authors report the case of a 36-year-old woman with CTS due to hibernoma of the wrist.

RÉSUMÉ

Le syndrome du canal carpien (SCC) est le plus fréquent des syndromes canalaires. Il est le plus souvent idiopathique, mais peut être causé par une lésion occupant l'espace intracanalaire, condition relativement rare voire exceptionnelle lorsque cette lésion correspond à une tumeur peu fréquente dont la localisation est atypique. Les auteurs rapportent le cas d'une patiente de 36 ans présentant un SCC secondaire à un hibernome du poignet.

1. Introduction

Carpal tunnel syndrome (CTS) due to space-occupying lesions is a rare or even exceptional entity in the case of a tumor [1]. We report here the case of a 36-year-old woman with CTS due to hibernoma.

2. Clinical case

A 36-year-old patient, right-handed, housewife, with no notable medical or surgical history, consulted for finger numbness of her right hand evolving over a year and gradually worsening.

The physical examination noted a painless mass of the anterior aspect of the wrist. This mass was movable with respect to the superficial plane, very limited, with a diameter of 1 cm and of firm consistency. Besides, the examination revealed a slight muscle atrophy of the thenar eminence in comparison with the opposite side. The neurological examination showed hypoesthesia in the median nerve territory. The vascular and muscular examinations were normal.

X-rays of the wrist were normal. Nonetheless, the ultrasound examination proved the presence of an oval-shaped mass of a diameter of 1.74 cm, with tissue echogenicity and clear limits, occupying the proximal edge of the carpal tunnel (Fig. 1). In addition, an electroneuromyogram (ENMG) revealed a moderate decrease in nerve conduction of the median nerve alongside with an increase in motor latency. As a result, the diagnosis of secondary carpal tunnel was considered and surgery was decided. Unfortunately, MRI could not be performed due to lack of availability.

The time period between the first consultation and the surgery was 6 weeks. After carrying out infraclavicular block and placing a tourniquet at the base of the limb, incision was made at the anterior aspect of the wrist. Surgical exploration found a limited tissue lesion in front of the median nerve at the superior border of

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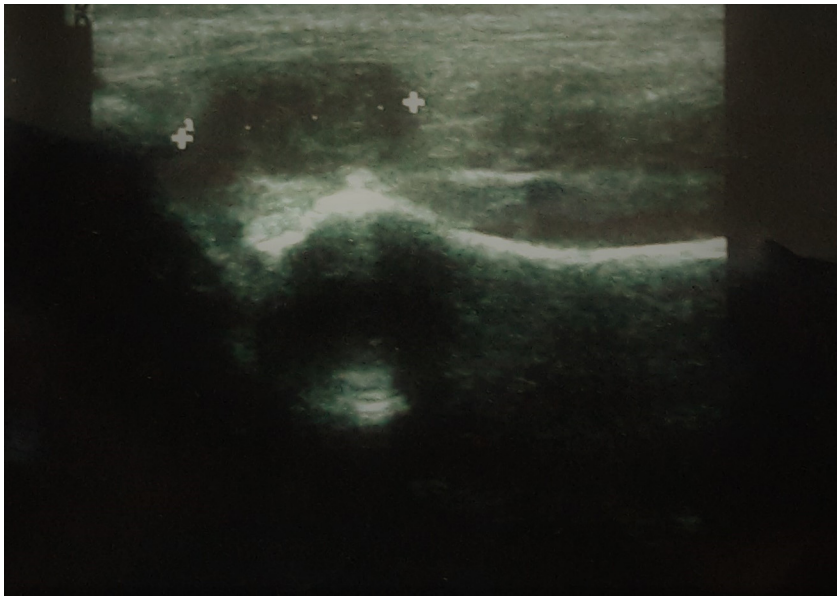

Fig. 1. Ultrasound image showing the appearance of the tissue lesion (between the two crosses).

the carpal tunnel (Fig. 2). Thus, the surgery consisted in the total resection of the mass. Finally, no associated muscle or tendon lesion was found.

During the follow-ups, the patient reported a regression of paresthesia from the 3rd week with complete disappearance by the end of the 6th week. By the last follow-up after 14 months, the patient was satisfied with no recurrence of the same symptomatology. The ENMG performed in the 8th month was normal.

Anatomo-pathological examination of the resected mass revealed the presence of monomorphic round cells with central nucleus and eosinophilic granular cytoplasm in favor of a brown fat tumor or hibernoma (Fig. 3).

3. Discussion

CTS due to space-occupying lesions is surely a rare entity, accounting for only 5.5% of all CTS [1]. Many authors have considered CTS due to tumors: lipoma [2], hemangioma [3], synovial sarcoma [4], and synovial giant cell tumor [5]. No article reports CTS due to hibernoma, which marks the originality of our observation.

Hibernoma is a slow growing benign tumor that develops at the expense of fat cells in brown fat. This tumor, described by Merkl in 1906, owes its name to Gery by analogy to the brown fat of hibernating animals [6]. Physiologically present in fetuses and newborns, brown fat accounts for only 1% of an adult body weight and is mainly located in the interscapular region and mediastinum [7].

Hibernoma is a rare tumor, accounting for only 1.6% of benign adipose tissue tumors [8]. It concerns most often young adults between 30 and 40 [1,7]. The predominance of sex is still controversial; some authors report male predominance [9] while others consider female predominance [7,10]. Several locations have been described in the upper limb: shoulder (11.8%), arm

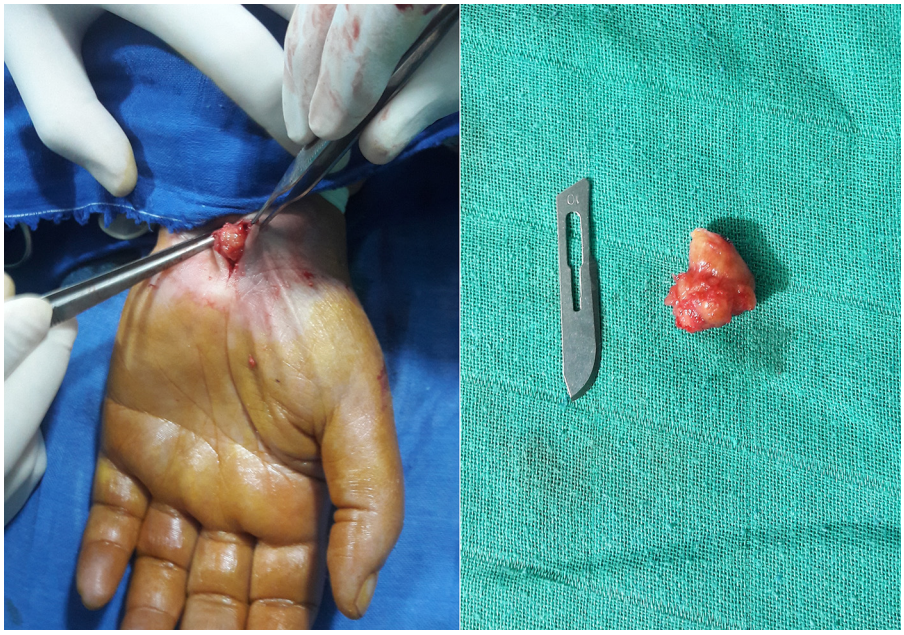

Fig. 2. Intraoperative image showing mass (a).The mass after excision (b).

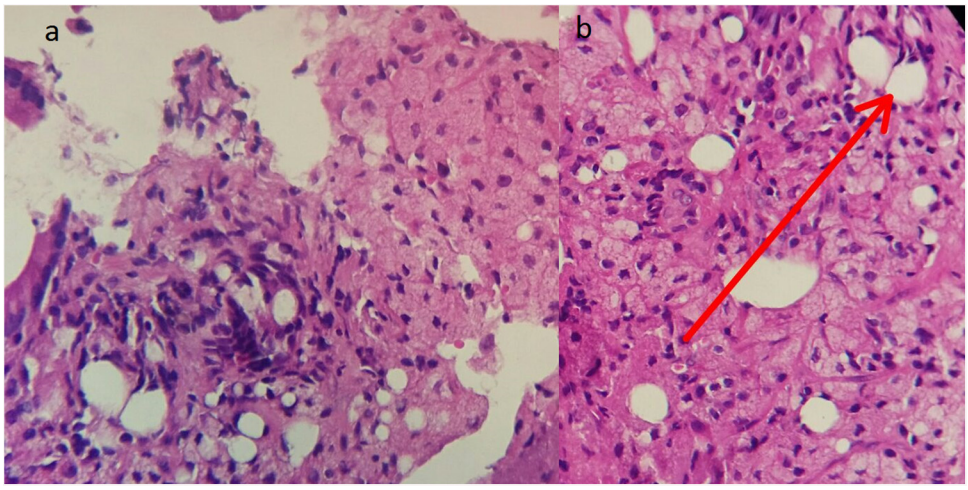

Fig. 3. Lobular tumor proliferation of microvacuolar eosinophilic cells (HE × 200) (a). Tumor cells with microvacuoles mixed with some adipocytes (red arrow) (HE × 400) (b).

(6.5%) [9], only one case reported in the forearm [11]. We found no case of hibernoma in the hand or wrist, which reinforces the particularity of our observation.

Clinically, CTS due to space-occupying lesion is considered when its symptomatology is unilateral. The tumoral origin is most suspected in case the symptoms evolve gradually without trigger factor and without period of remission with permanent paresthesia not having the usual nocturnal predominance [12]. However, the unilateral character is not specific to CTS due to space-occupying lesion; indeed, bilateral CTS may not be symptomatic on one side and it is described to be "infra-clinical", which justifies the importance of carrying out bilateral and comparative examination during the ENMG [1]. Hibernoma is clinically manifested only by pain due to neighboring organ compression or by exceptional weight loss in relation to excessive metabolic activity of the tumor [7].

Medical imaging is not one of the usual means of exploration for CTS. However, it is essential for eliminating CTS due to space-occupying lesions when it is unilateral and the etiology is unclear, based on medical history and physical examination [1]. Medical imaging is also important in the diagnosis of hibernoma which appears in Doppler ultrasound as a highly vascularized homogeneous echo-structure and represented in CT scan with fat density [7]. In MRI, the signal intensity of hibernoma is between that of skeletal muscle and that of subcutaneous fat: T1 signal is lower than that of subcutaneous fat with higher T2 signal [13].

Confirmatory diagnosis is anatomopathological: histologically, these tumors show a lobulated architecture, composed of oval or round monomorphic cells, with small central nucleus and eosinophilic granular or multivacuolated cytoplasm [11]. A histological classification is based on the nature of the stroma and the appearance of multivacuolated cells: the typical form, the lipoma-like tumor, the myxoid form and hibernoma with spindle-shaped cells [10]. The blood flow is generally more abundant than in a lipoma [11]. In immunohistochemistry, these tumors are positive for PS100 and CD34. Alterations in the q13 region of chromosome 11 on a locus close to the gene of multiple endocrine neoplasia type 1 (*MEN 1*) have been incriminated [14].

Surgical resection is essential for treatment but also for histological confirmation [11]; thus, liposarcoma is the main differential diagnosis and is eliminated only after anatomopathological examination of the resected tumor. The risk of degeneration of the hibernoma is disputed [10], which justifies the importance of complete resection.

4. Conclusion

The CTS due to tumor lesion is a rare entity that must be considered when the nature of the paresthesia is unilateral and permanent, and must be confirmed by the ENMG and sought by imaging.

Disclosure of interest

The authors declare that they have no competing interest.

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