Clinics in Surgery

Case Report
Published: 18 Jul, 2018

Rare Founding in Peripheral Nerve Surgery: An Unicentric Castleman Disease Presenting as Median Nerve Tumor: Case Report

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Abstract

A 51 year old man presented with progressive swelling in the upper arm. MRI revealed a solitary mass extending from the median nerve. Intraoperative finding was a tumor extending within the nerve in its proximal fibers. The histological result showed typical criteria of a Castleman disease. Postoperative staging did not reveal any involvement of other organs. In conclusion, an unicentric Castleman disease could be diagnosed. To our knowledge such an entity appearing at a peripheral nerve has not been described in literature so far.

Keywords: Peripheral nerve tumor; Unicentric Castleman disease; Contrast enhancing mass; Infiltrative growth; Surgery

Introduction

Tumors of the peripheral nerve system represent a broad and inhomogeneous group. They can be subdivided into two categories according to their intrinsic or extrinsic growth: Peripheral nerve sheath tumors (PNST) and Peripheral non-neural sheath tumors (PNNST). Each entity includes benign and malignant tumors [1,2] (Table 1). Diseases arising from lymphoid cells which are associated to the peripheral nerve system are extremely rare. A few cases of primary lymphoma [3], neurolymphomatosis [4] and lymph node metastasis [5] have been described previously. We present another entity of lymphoid tissue found in a peripheral nerve. It is known as Castleman diseaese (CM) or angiofollicular lymph node hyperplasia [6].

Case Presentation

A 51 year old man presented to our outpatient department with an increasing swelling in the right distal upper arm. He reported about local pain without radiation. There was no loss of sensory or motor function. The patient´s medical history was empty, no previous infections, surgeries or other diseases. The mass in the arm presented solid and relocatable.

MRI of the upper arm showed a spindle-shaped homogeneously contrast enhancing mass. Its diameter was about 11 cm × 4 cm. It was located in the course of the median nerve respectively seemed to originate from part of its fibers (Figure 1a and 1b). The primary diagnosis from the radiologist was schwannoma.

Surgical extirpation was indicated and performed. In its middle part the exposed tumor had a smooth capsule which was opened (Figure 2a). In its equator the surface had a good boundary to the surrounding tissue (Figure 2b). It did not extend to the muscles or tendons. In its distal and especially in its proximal ending the tumor showed a more infiltrative growth (Figure 2c). A feeding fascicle could be identified and was cut after ensuring by electric stimulation that it had no motor function. But with the intention to set no damage at the main nerve trunk approximately twenty percent residual tumor was left (Figure 2d).

The postoperative course was uneventful. The patient suffered a light hypoesthesia in the forearm. This did not match to the supply territory of the median nerve which is the palmar hand. It rather corresponded to another skin nerve, possibly damaged by the approach. The local upper arm

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Received Date: *20 Jun 2018*
Accepted Date: *13 Jul 2018*
Published Date: *18 Jul 2018*

Citation:
Carolus A, Schroers R, Tischoff I, Schmieder K, Brenke C. Rare Founding in Peripheral Nerve Surgery: An Unicentric Castleman Disease Presenting as Median Nerve Tumor: Case Report. Clin Surg. 2018; 3: 2035

Table 1: Tumors of the peripheral nerve system.

MPNST	BPNST	MPNNST	BPNNST
MPNST	- schwannomas (= neurilemmomas/ neuinomas) - neurofibromas	- metastasis (lung, breast, melanoma) - sarcomas - lymphomas	- ganglioncysts - hypertrophic neuropathy - lipomas - venous angiomas - hemangiopericytomas - hemangioblastomas - myositis ossificans - ostochondromas - ganglioneuromas - meningeomas - cystic hygromas - myoblastomas - granular cell tumors - epidermoid cysts - desmoids

MPNST: Malignant Peripheral Nerve Sheath Tumour; BPNST: Benign Peripheral Nerve Sheath Tumour; MPNNST: Malignant Peripheral Non Neural Sheath Tumour; BPNNST: Benign Peripheral Non Neural Sheath Tumour

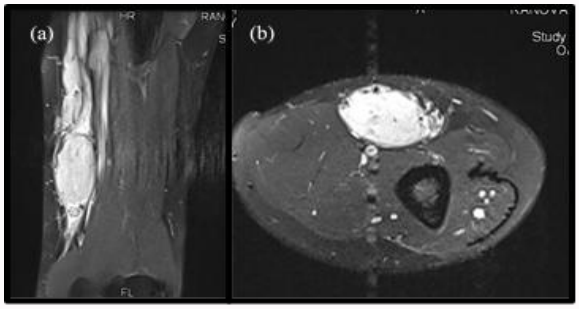

Figure 1: MRI of the upper arm showing a spindle shaped contrast enhancing mass in the median nerve course. a) Coronar view. b) Axial view.

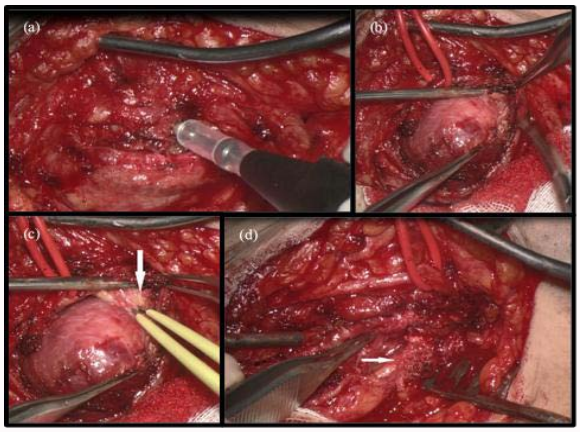

Figure 2: Intraoperative views of tumour dissection and removal. a) Opening the capsule of the tumour and electric stimulation of fascicle-like structures. b) Exposed tumour with a smooth surface in its middle part. c) Exposed tumour, infiltrative growing in its proximal ending (arrow). d) Tumorbed after removal of the median part, rest tumour embedding a nerve branch (arrow).

The final histological examination of the tumor showed typical criteria of the Castleman disease with an effaced architecture of a lymph node with regressed germinal centers and typical high endothelial venules (Figure 3a and 3b). Immunohistochemistry demonstrated regressed atrophic germinal centers (Figure 3c) and aberrant network of follicular dendritic cells (Figure 3d). The combination of these features ensured the diagnosis.

To exclude a multicentric disease the patient was admitted to the internal medical department. Entire virus tests including HIV were negative. A bone marrow biopsy showed a normal hematopoiesis

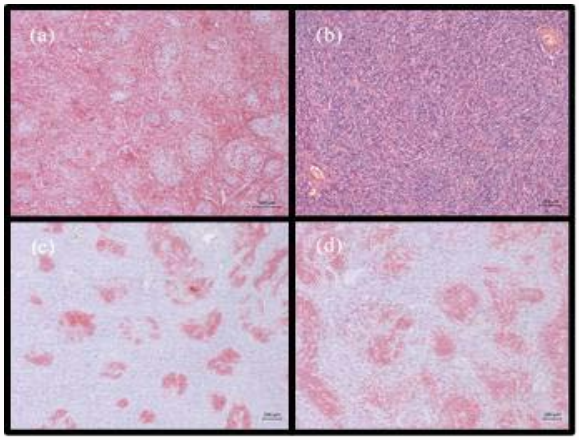

Figure 3: Histological stain sections. a) Immunhistochemical staining for T-cells within the interfollicular zone. b) In 10x HE stain section areas with high endothelial venules are demonstrated. c) Immunhistochemical expression of CD20 demonstrating regressed germinal centers. d) Immunhistochemical expression of CD23 shows extended network of follicular dendritic cells.

without evidence for an infiltration by pathologic cells. A staging PET-CT showed no further organ manifestations. An unicentric form was approved in synopsis of all findings. In regard to the tumor rest and the curative approach of a unicentric M. Castleman the patient finally underwent a selective radiation of the upper arm [7]. In a 6 month follow-up the patient reported no nerve related problems or restrictions in everyday life.

Discussion

Retrospectively one can discuss what would have been the best neurosurgical management for our patient. Would he have benefit from a more radical tumor resection? Or contrariwise, would a frozen section have been helpful in such a case and a biopsy been the consequence? Should staging have had priority?

According to the literature unicentric Castleman disease has a good prognosis concerning the overall survival if treated by either complete surgical removal or a combination of surgery and radiation [7]. In this respect we think that an extirpation should out value a sole biopsy in such a case. But peripheral nerve function preservation has priority, especially if a major nerve of the upper or lower extremity is involved.

The histological dignity and growth behavior of a peripheral nerve tumor is irremissible information in this benefit-risk-assessment

between complete removal on the one side and preservation of sensorimotor function on the other side. The features from Castleman disease distinguish clearly from the characteristic of tumors originating from the nerve sheath which is neoplastic proliferation of Schwann cell differentiation [8]. Nevertheless there is an increasing literature of MPNST mimics. Concerning the MRI finding and the infiltrative growth our case represents not a microscopic, but a macroscopic differential diagnosis of MPNST.

In our opinion the fact of its rareness, the challenging MRI, the malignant histological features and the necessity of interdisciplinary treatment justify that unicentric Castleman disease is registered in the itemization of peripheral nerve-associated malignant neoplasms.

Conclusion

Castleman disease is a group of lymph proliferative disorders including special histological characteristics of lymph node. It is rare and may affect any body region. A manifestation at a solitary peripheral nerve has not been described so far. We present a case in which an unicentric Castleman disease mimicked a MPNST if dealing with the radiological and neurosurgical features. The pathological examination resolved the challenging case. An interdisciplinary treatment approach seems to be essential.

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