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Obturator mononeuropathy due to adenocarcinoma of unknown primary origin

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Summary

A 35-year-old woman presented with a three-month history of left groin and thigh pain. Neurological examination and electromyography showed pathological features consistent with obturator nerve involvement. Imaging studies revealed a left retroperitoneal mass, which by pathological examination was shown to be metastatic adenocarcinoma of possible Müllerian origin. Primary tumor could not be detected in a follow-up period of three years. Obturator mononeuropathy can be the first manifestation of cancer. Cancer of unknown primary origin may occasionally be local, well-restricted and carry a good prognosis.

Key words: Obturator nerve; Mononeuropathy; Groin pain; Tumor; Adenocarcinoma.

Introduction

Obturator mononeuropathy is a rare condition usually caused by trauma, surgery or lithotomy positioning, whereas obturator nerve compression due to tumors might also rarely occur. In the latter case, usually a metastatic tumor entraps the nerve in or before the obturator canal leading to well-characterized motor and sensory symptoms [1-3].

Case Report

A 35-year-old woman with no history of recent trauma or surgery presented with a three-month history of increasing pain in the left groin and thigh. Neurological examination showed reduced adductor muscle strength (4/5 by medical research council scale) in the left leg and diminished pinprick sensation along the medial left thigh and was otherwise normal. The groin pain was exacerbated by extension of the left leg and there was no color change or edema in the legs. Needle electromyography examination showed denervation findings (fibrillation potentials, high amplitude, long duration complex motor unit potentials) in the adductor muscles of the left leg, whereas other limb muscles and paraspinal muscles innervated by L2-4 nerve roots, which also supply the obturator nerve, were spared. Nerve conduction studies were normal. Magnetic resonance imaging showed a retroperitoneal mass located on the lateral side of the left internal iliac artery. A positron emission tomography (PET) scan did not show any additional lesions. Following the surgical resection, the pathological examination identified the mass lesion as poorly differentiated adenocarcinoma of unknown primary origin. The tumor tissue contained extensive regions of necrosis and tumor cells with vesicular nucleus, distinct nucleolus, and wide eosinophilic or clear cytoplasm (Figure 1). An extensive immunohistochemical screening revealed tumor cells that were positive for paired box gene 8 (PAX8), estrogen receptor, cytokeratin 7 and vimentin, and negative for p53, cytokeratin 20, Wilm's tumor suppressor gene1 (WT1), CD10, renal cell carcinoma marker (RCC Ma), thyroid transcription factor 1 (TTF1), synaptophysin, chromogranin, and T-cell-specific transcription factor GATA-3. Tissue samples obtained from bladder, endometrium, and cervical canal did not contain any tumor cells. Only radiotherapy was performed due to absence of clinical and pathological evidence for local tumor invasion. After the treatment, the patient's complaints completely regressed and did not recur in a follow-up period of three years. Despite several repeat PET studies, the primary tumor could not be detected in this timeframe.

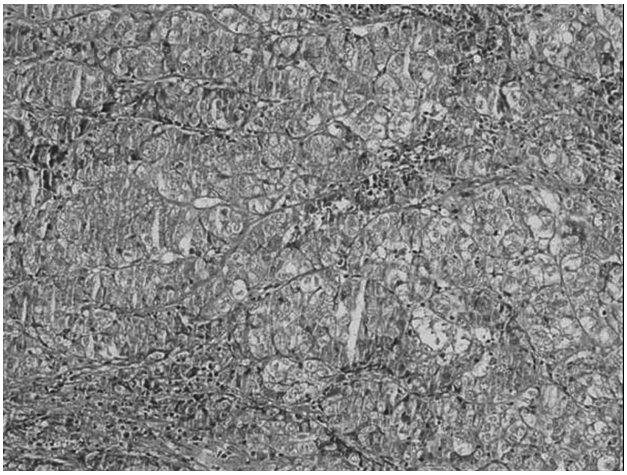

Figure 1. — Histopathologic image of the carcinoma of unidentified primary origin. Tumor cells have vesicular nucleus, distinct nucleolus, and wide eosinophilic or clear cytoplasm (hematoxylin and eosin stain, magnification x200).

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Discussion

Persistent groin pain can be produced by a variety of conditions including tendinitis, bursitis, hernia, and inflammation or fracture of the pelvic bones [2, 3]. Although entrapment of the obturator nerve especially by a tumor is a relatively rare cause of groin pain, the present case reinforces the notion that tumor screening should be performed in patients with obturator neuropathy findings and no history of trauma or surgery, especially because obturator mononeuropathy might often be the sole presenting sign of a pelvic or retroperitoneal tumor.

After descending through the major psoas muscle, the obturator nerve proceeds along iliac arteries and ureter and enters the thigh through the obturator canal, where it divides into anterior and posterior branches. In a few reported obturator mononeuropathy cases caused by direct impingement or infiltration of a tumor, major sites of nerve injury were obturator canal or pelvis. Moreover, the detected tumors were almost invariably malignant cancers, mostly originating from pelvic or abdominal tissues such as bladder, prostate, and kidney [1, 2]. Although there is a single reported obturator neuropathy case caused by a cancer of unknown primary origin (CUP), detailed pathological features and long-term prognosis of this case have not been provided [1].

CUP is a common entity that accounts for 3-5% of malignant epithelial tumours and only 20% of CUP cases have a favorable prognosis. In around 2-5% of CUP cases, the primary site is never identified. Pathological diagnosis in the majority of CUP cases is adenocarcinoma and only in around one-third of these cases the tumor is poorly differentiated [4, 5]. Although origin of the CUP is estimated through an immunohistochemistry screening for prognosis and treatment purposes, tumor cells of the present patient did not express most of the molecules produced by metastatic adenocarcinomas. However, detection of PAX8, which is primarily expressed by thyroid, thymus, renal cell cancers, and carcinomas of Müllerian origin, was a key finding [6]. Absence of TTF1 and RCC Ma staining in the tumor tissue largely eliminated thyroid and renal cancer possibilities [7, 8]. Failure to detect any tumor activity in thymus or other tissues by repeat PET examinations in a time span of three years and presence of PAX8 staining suggested that the tumor causing obturator neuropathy in the present case was presumably a local retroperitoneal neoplasm originating from the Müllerian duct.

Obturator mononeuropathy can be the first manifestation of cancer and thus should be considered in patients with groin pain. Although CUP usually has a metastatic and aggressive nature, it may also occasionally be local, well-restricted, and carry a good prognosis, as observed in the present case.

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