



Malignant Glomus Tumour with A Rare Localization

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ABSTRACT

Glomus tumor is a mesenchymal tumor that results from differentiation of modified smooth muscle cells responsible of thermoregulation. They account for <2% of soft tissue tumors. Although these tumors are found in all body parts, 80% of them are placed in the upper extremities and 75% of them are located beneath finger nails. Malignant glomus tumor, which mostly shows local infiltration, is very rare accounting for 1% of total glomus tumors. Our case, which describes a malignant glomus tumor localized in patella, is presented and discussed because of its rare location and posttreatment recurrence

Keywords: Glomus tumor, patella, recurrence

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INTRODUCTION

Glomus tumor is a mesenchymal tumor comprising smooth muscle cells known as glomus cells; it is responsible for thermoregulation (1). It was first described by Masson in 1924 (2). Glomus tumors constitute <2% of soft tissue tumors (3). They are seen more commonly in the fourth and fifth decades of life in all age groups. Clinically, they are usually red and bluish-purple lesions and are <1 cm in size. Cold sensitivity, point sensibility, and paroxysmal pain are the most important features of these tumors (4). Glomus tumors, which usually show a subungual location, are rarely found outside the finger; however, cases have been reported in the foot, ankle, knee, hip, thigh, thorax, sacrum, and coccyx (5). A malignant glomus tumor is seen very rarely. It constitutes 1% of all glomus tumors and usually progresses with local infiltration (6, 7). Post-surgery local recurrence has generally been reported as 10% and is usually attributed to poor excision (8). In this article, a case of patellar malignant glomus tumor was presented due to the fact that it had a rare localization and recurred after treatment.

CASE REPORT

A 31-year-old male consulted the clinic due to pain in his right knee. Open biopsy performed for the same complaints two years ago reported "fusiform cell mesenchymal tumor," and resection and patellar reconstruction operation performed two months later revealed "lymphovascular proliferation, inflammatory granulation tissue. No residual tumor was seen, no tumor was seen at the surgical margins." At his subsequent application to the clinic, physical examination revealed pain after 110° flexion and pal-

pable tenderness in the right knee, and a mass was seen at the same location. Excisional biopsy revealed a macroscopic mass sized 2×3 cm. Microscopic examination of the mass revealed tumor cell islands that were surrounded by vascular structures in various sizes, had eosinophilic cytoplasm, oval-round nucleus, wide and single nucleoli, and polygonal and oval appearance. These cells contain cytologic atypia and mitotic activity (Figure 1). Positive reaction was observed in tumor cells with immunohistochemically applied vimentin and smooth muscle actin; however, no reaction was observed in tumor cells with pancytokeratin, carcinoembryonic antigen, and desmin (Figure 2). The case was evaluated in favor of malignant glomus tumor in the presence of clinical, morphological, and immunohistochemical findings. Informed consent was obtained from the patient for the publication of this case report.

DISCUSSION

Glomus tumors constitute a small proportion of soft tissue tumors; they can be solitary or multicentric, and are quite rare. Glomus tumors, which usually show a subungual location, are rarely found outside the finger; however, cases have been reported in the foot, ankle, knee, hip, thigh, thorax, sacrum, and coccyx (5, 9). A glomus tumor located in the anterior region of the patella has been reported with two cases in the literature (8). The case we present here also has a patellar localization; it is rarely seen and shows local recurrence. Local recurrence after surgery was generally reported as 10%; it is usually attributed to inadequate excision. Complete surgical resection was performed in our case. After resection, granulation tissue was observed at the same location. However, the patient had recur-

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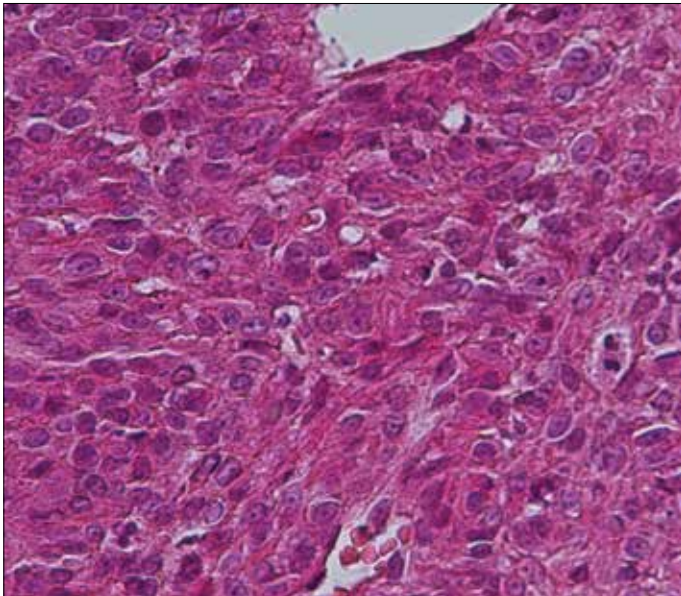


Figure 1. Presence of pleomorphism, atypia, large nucleolus, and mitotic potential (H&E; x400)

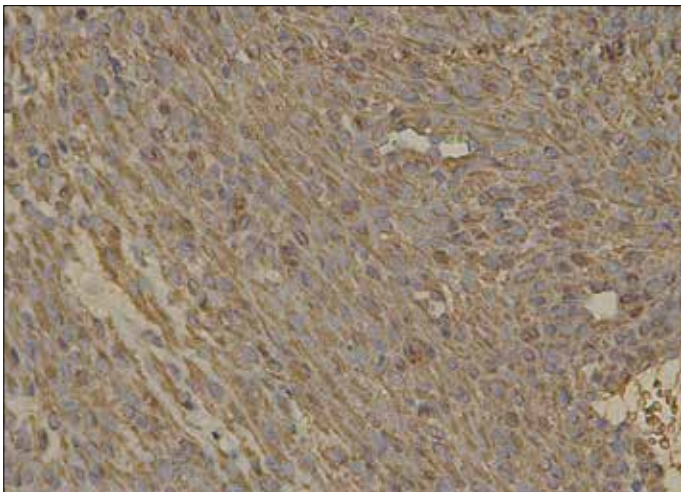


Figure 2. Cytoplasmic positive reaction is observed with immunohistochemically applied SMA (H&E; x200)

rent lesions in the same location in the examinations performed two years later. Putti and Tatò (8) reported that local recurrence developed after local excision in two cases with glomus tumors located in the anterior region of the patella, and they attributed this situation to the insufficient excision of the tumor. Contrarily, in our case, recurrence did not develop after local excision; however, it developed despite the fact that wide excision was performed and no tumor was seen at the surgical margin. Folpe et al. (7) defined the criteria for malignant glomus tumors as follows: deeply-located tumor, diameter >2 cm, presence of marked atypia along with atypical mitotic figures, and mitotic

activity. The presence of these characteristics has been found to be associated with the risk of metastasis. The presence of atypical mitosis revealed via the microscopic examination of our case overlaps with the malignancy criteria, i.e., atypia in tumor-forming cells and macroscopic diameter >2 cm.

CONCLUSION

Glomus tumors may rarely develop in regions outside the finger; it may be difficult to diagnose its involvements in these regions, and in cases of unexplained pain, glomus tumors should be considered in the differential diagnosis. Follow-up is required in terms of recurrence. In tumors sized >2 cm, malignant glomus tumors should be considered for differential diagnosis in the presence of nuclear atypia and atypical mitosis and should be followed up more closely in terms of local recurrence. It should also be considered that these tumors may have metastatic potential.

Informed Consent: Verbal informed consent was obtained from patient who participated in this case.

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