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Paraganglioma in paratesticular: A rare case report

Ahmed Mousa Almuhanna, Basim Alghorairy, Turki H Alessawi, Sara Sameer Albagshi, Abdulrahman Alhazeem, Hussain M AlModhi

ABSTRACT

Paraganglioma at a paratesticular location is extremely rare. We report a 58-year-old Saudi male presented with two years history of right painless scrotal mass. On physical examination the scrotum revealed a right-sided non-tender mass not attached to right testis. Normal tumor markers of testicular tumor. Ultrasonography revealed a well-defined, homogeneous, hyperechoic lesion measuring approximately 2 cm in the right extratesticular region. Magnetic resonance imaging (MRI) with intravenous (IV) gadolinium contrast for abdominal and pelvis showed right extratesticular soft tissue mass not separable from the spermatic cord and there was no distant metastasis. The patient underwent exploratory excision of the mass with preservation of cord and testis. Histopathology showed paratesticular paraganglioma.

Keywords: Paraganglioma, Testicular, Tumor

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INTRODUCTION

Paragangliomas are unique rare neuroendocrine neoplasms arising from neural crest [1]. These tumors are usually located in the carotid body, the jugulotympanic body, or mediastinal vessels with the majority being benign. Although they have been described in most organ but rarely found in genital site, a paraganglioma at a paratesticular location is extremely rare. The clinical features of these tumors are variable, including hypertension, palpitations, headache, sweating, and other symptoms associated with increased catecholamine levels [1]. We report a case of asymptomatic painless solitary right primary paraganglioma in the paratesticular region.

CASE REPORT

A 58-years-old Saudi male presented with two years history of right painless scrotal mass. There was no history of fever, genital trauma, genital infection, or tuberculosis (TB). The patient denied having nausea, vomiting, diarrhea, flushing, palpitations, or weight changes. On physical examination, the scrotum revealed a right-sided non-tender mass with 2 cm above and not attached to right testis. The left side of the scrotum was normal and no palpable lymph nodes. Normal tumor markers of testicular tumor. Ultrasonography revealed a well-defined, homogeneous, hyperechoic lesion measuring approximately 2 cm in the right extratesticular region.. It was seen superior to right testis and right epididymis and separable from them with high vascularity by Doppler study. Both testes appeared average in size and normal vascularity (Figure 1). Magnetic resonance imaging (MRI) with intravenous (IV) gadolinium contrast for abdominal and pelvis showed right extratesticular soft tissue mass not separable from the spermatic cord and there was no distant metastasis (Figure 2A–C). Based on images, provisional clinical diagnosis of a benign paratesticular

mass was made and the patient underwent exploratory excision of the mass with preservation of cord and testis. The patient was discharged two days after surgery. Histopathology showed grossly a single soft gray light brown tissue mass measuring 2×2 cm with white firm focally fleshy cut surface. Microscopically, cells arranged in nested and trabecular growth pattern. The neoplasm is composed of tumor nests comprising of round cells with abundant granular eosinophilic-basophilic focally clear cytoplasm surrounded by sustentacular cells embedded in vascular rich stroma. The lesion exhibited no evidence of capsular invasion, lymphovascular invasion, a diffuse pattern, tumor necrosis, atypical mitosis, or increased mitotic activity. Histopathology reported this mass is paraganglioma.

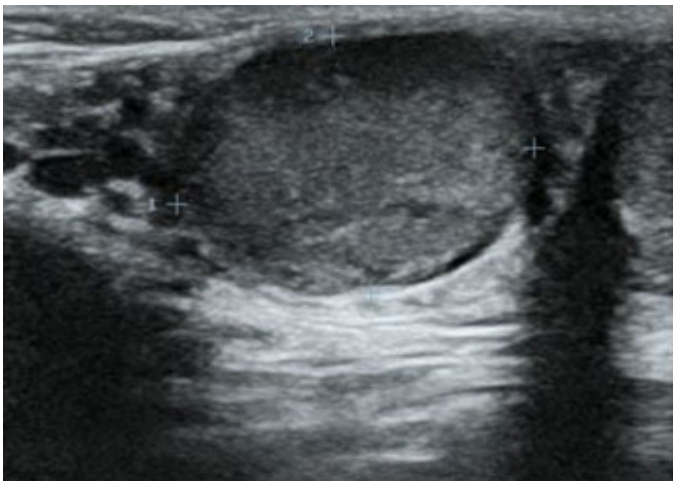

Figure 1: Ultrasonography showed homogeneous hyperechoic lesion measuring approximately 2 cm.

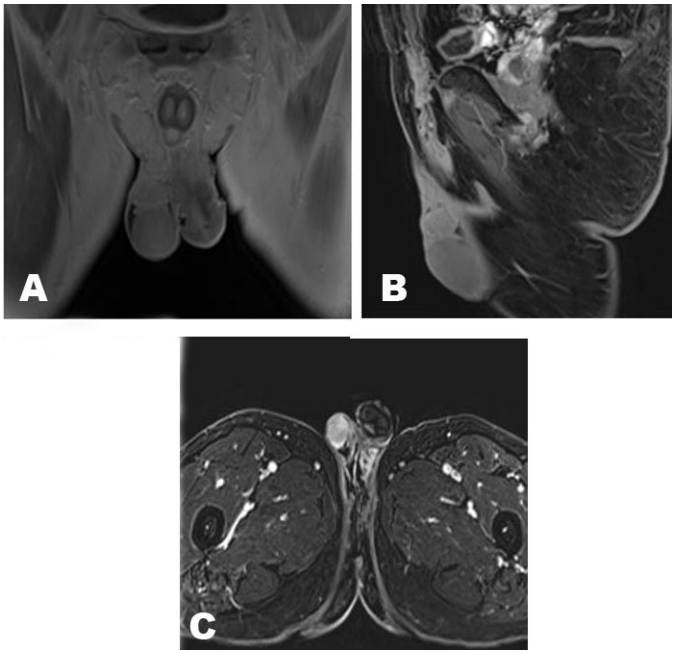

Figure 2: (A–C) MRI showed right extratesticular soft tissue mass not separable from the spermatic cord.

DISCUSSION

Scrotal paratesticular masses are diagnostic dilemma. About 30% of paratesticular tumors are malignant. Therefore, all scrotal masses should be adequately evaluated [2]. Exploratory surgery is the definitive treatment. If mass is benign, a testis-sparing surgical procedure can be performed. Common benign tumors of the paratesticular region include lipomas, adenomatoid tumors, leiomyomas, fibromas, papillary, and cystadenomas [2]. But paragangliomas are extraordinarily rare tumors at this site. Generally, paragangliomas are neuroendocrine neoplasms of neural crest. They occur in the fourth and fifth decades and are equally prevalent in both genders [3]. Most paragangliomas are located in the adrenal gland, where they are referred to as pheochromocytomas. Extra-adrenal paragangliomas are found along the sympathetic and parasympathetic chains. The common sites for extra-adrenal paragangliomas include the carotid body, vagal body, middle ear, abdomen, and laryngeal areas [2]. Paragangliomas have occasionally been reported in the urogenital tract, in the urinary bladder [4], and the prostate [5]. The first reported case of paratesticular paraganglioma was found in 1971 by Eusebi et al. [6]. There are only few cases have been reported worldwide in the literature [7]. Paraganglioma can be functional or non-functional. In a functional lesion, the tumor secretes catecholamines and the presenting symptoms, such as headache, sweating, palpitations, and hypertension, are secondary to elevated levels of catecholamines. Our patient was asymptomatic and was not tested for catecholamine levels prior to surgery. Most paragangliomas are benign. Malignant lesions are rare occur in the fifth to seventh decade, and they tend to be more symptomatic than benign tumors. The diagnosis of malignancy is essentially based on the presence of distant metastasis. The lesion displays capsular or lymphovascular invasion, a diffuse pattern, tumor necrosis, atypical mitosis, increased mitotic activity, and protein-positive sustentacular cells. These findings indicate for malignancy [3]. However, findings are not reliable to establish diagnosis of malignancy. Therefore, the treatment of choice for a paraganglioma is complete surgical resection with regular follow-up to rule out a recurrence or metastases. Our patient underwent excision of right paratesticular paraganglioma and we follow him up for six months with physical examination only, since there is no clear protocol for follow-up.

CONCLUSION

Primary paratesticular paragangliomas are very rare. We believe that despite their rare incidence, they should be considered in the differential diagnosis of paratesticular tumors. Neither radiological imaging is sufficient to determine the nature of a paratesticular mass nor laboratory workup. The definitive diagnostic

modality is surgical exploration and excisional biopsy for histopathological evaluation.

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Author Contributions

Ahmed Mousa Almuhanna – Conception of the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Basim Alghorairy – Conception of the work, Analysis of data, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Turki H Alessawi – Design of the work, Interpretation of data, Drafting the work, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Sara Sameer Albagshi – Interpretation of data, Drafting the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Abdulrahman Alhazeem – Analysis of data, Interpretation of data, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Hussain M AlModhi – Acquisition of data, Interpretation of data, Drafting the work, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

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Conflict of Interest

Authors declare no conflict of interest.

Data Availability

All relevant data are within the paper and its Supporting Information files.