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Accidentally discovered adolescent botryoid rhabdomyosarcoma

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INTRODUCTION

Rhabdomyosarcoma (RMS) is a rare malignant tumor arises from the embryonic muscles of the female genital tract of children, and adolescents, and it accounts for 4-6% of all malignancies in childhood [1].

The botryoid RMS is a polypoid form of RMS with a "grape-like" appearance. The origin of botryoid RMS is closely related to the age; it's originated from the vagina in infancy, and childhood, from the cervix in reproductive-age, and from the uterine corpus in postmenopausal women [2].

The botryoid RMS of the vagina is more common than the cervical botryoid RMS, while the cervical botryoid RMS has better prognosis [2]. The botryoid RMS tends to recur locally after excision and invades the adjacent organs [2].

The management of botryoid RMS is challenging especially at a young age (*i.e.*, childhood, or adolescents), as the preserved reproductive and sexual functions are essential.

There are no uniform consensus and/or guidelines regarding the surgical, and/or the conservative treatment of botryoid RMS.

Therefore, this report represents an adolescent botryoid RMS, to highlight the management challenges of botryoid RMS.

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A 13-years-old adolescent girl, admitted to the hospital because of iron deficiency anemia (IDA) caused by the abnormal uterine bleeding (AUB). She had a normal hormonal profile (TSH 2.5 mIU/mL, prolactin 195 mIU/mL, FSH 5.3 mIU/mL, and LH 3.7 mIU/mL), normal liver enzymes (ALT 22 U/L, and AST 19.5 U/L), normal APTT 28 Sec., normal bleeding 3.8. min., and clotting 4.8 min. times.

The trans-abdominal sonography (TAS) showed a normal uterus, without any intrauterine lesions, and normal both ovaries.

The patient hemoglobin was 9.2 g/dL, and she received intravenous iron saccharate/sucrose (Spimaco, Saudi Arabia) to correct the IDA (ferritin was < 15 µg/L), and intravenous tranexamic acid (Advanz pharm., United Kingdom) to control the AUB.

The PALM (defines structural AUB; polyp, adenomyosis, leiomyoma, and malignancy), and COEIN (defines functional AUB; coagulopathy, ovulatory, endometrial, iatrogenic, and non-classified) classification of AUB facilitates the AUB diagnosis, and management.

During admission, the patient`s passed a fleshy, reddish "Grapelike" polypoidal lesion through the external genitalia. The "Grapelike" lesion sent for histological examination, and unfortunately the diagnosis of botryoid RMS was confirmed by two senior pathologists. The histological examination showed a non-keratinizing squamous epithelium with an edematous stroma underlying, plump stromal cells with dense eosinophilic cytoplasm, and mitotic activity (Fig. 1).

After control of the AUB, a departmental imaging/metastatic work-up, including pelvic magnetic resonance imaging (MRI), computerized tomography (CT) of the thorax, and bone scan was done, followed by referral of the patient to the specialized oncology center. Departmental approval, and written consent were obtained from the studied adolescent`s parents to publish the studied adolescent`s data as a clinical vignette.

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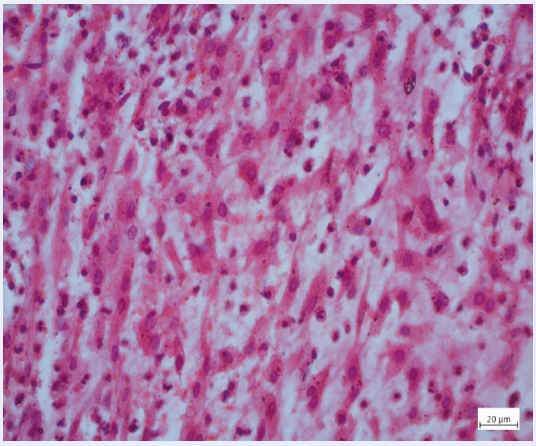

Figure 1. Botryoid (embryonic) rhabdomyosarcoma; embryonic myoblasts in myxoid stroma (staining with hematoxylin and eosin, magnification 400

DISCUSSION

This report represents a case of botryoid RMS presented with dysmenorrhea, and AUB, to highlight the management challenges of botryoid RMS.

The botryoid RMS is a rare malignant tumor, and the published data regarding the botryoid RMS are available from the published case-series, and case-reports [1].

The largest published series of botryoid RMS of the uterine cervix include 13 cases [3]. The botryoid RMS accounts for 10% of all RMSs. The botryoid RMS of the cervix tends to occur at older ages (i.e., children and adulthood), than those occur in the vagina [1].

Most of the botryoid RMS cases presented with either AUB and/or protruding mass from the introitus [2]. The differential diagnosis of botryoid RMS includes, adenosarcoma, cervical mesodermal polyp, and rhabdomyoma [1].

Complete surgical resection of botryoid RMS with a "safety margin" of normal tissue around is less applicable in metastatic botryoid RMS [1].

Daya and Scully [4], reported comparable results in 3 cases of botryoid RMS (out of 13 cases) treated using the fertility-sparing approach (*i.e.*, trachelectomy or polypectomy) with postoperative chemotherapy to those treated with radical surgery. This suggests that the cervical botryoid RMS may have a favourable outcome than those originated from the vagina [1].

Deletion of chromosome 1 short arm, and trisomies 13 and 18 were reported in RMS of uterine cervix [5].

CONCLUSIONS

Botryoid RMS is a polypoid form of RMS with a "grape-like" appearance. The botryoid RMS of the vagina is more common than the cervical botryoid RMS, while the cervical botryoid RMS has better prognosis. There are no uniform consensus and/or guidelines regarding the surgical, and/or the conservative treatment of botryoid RMS. Further studies evaluating the outcome of different management strategies of botryoid RMS are needed. An international guideline for the management of botryoid RMS are also needed.

Article informations and declarations

Ethics statement

Departmental approval, and written consent were obtained from the studied adolescent`s parents to publish the studied adolescent`s data as a clinical vignette.

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Conflict of interest

Authors declare no conflict of interest related to this clinical vignette.

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