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WORD COUNT	2175	TIME SUBMITTED	28-JAN-2023 09:50AM
		PAPER ID	95981079

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Name of Journal: *World Journal of Clinical Cases*

Manuscript NO: 82044

Manuscript Type: CASE REPORT

Primary yolk sac tumor in the abdominal wall in a 20-year-old woman: a case report

Primary YST in the abdominal wall

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Abstract

BACKGROUND

Extragonadal yolk sac tumours are rare, with only a low reported tumour occurrence outside the gonads locally and abroad. Extragonadal yolk sac tumour are usually a diagnostic challenge, because they are infrequent, but also because a thoughtful and detailed differential diagnostic process must be performed.

CASE SUMMARY

Here we present a case of an abdominal wall yolk sac tumour in a 20-year-old woman admitted with a tumour in the lower abdomen close to the umbilicus. The tumorectomy was performed. The histological examination revealed characteristic findings such as Schiller-Duval bodies, loose reticular structures, papillary structures, and eosinophilic globules. According to the immunohistochemical staining, the tumour tissue was positive for broad-spectrum cytokeratin, Sal-like protein 4, glypican-3, CD117, and epithelial membrane antigen（EMA）. Based on the clinical information,

histological features, and immunohistochemical staining profile, the tumour was diagnosed as a yolk sac tumour present in the abdominal wall.

CONCLUSION

Based on the clinical information, histological features, and immunohistochemical staining profile described above, the tumour was diagnosed as a primary yolk sac tumour in the abdominal wall.

Key Words: Yolk sac tumor; abdominal wall; 20-year-old woman; case report

Wang Y, Yang J. Primary yolk sac tumor in the abdominal wall in a 20-year-old woman: a case report and review of literature. *World J Clin Cases* 2023; In press

Core Tip: Extragonadal yolk sac tumor are usually a diagnostic challenge, because they are infrequent, but also because a thoughtful and detailed differential diagnostic process must be performed. In this case, since the tumor presented classic yolk sac tumor features, and immunohistochemistry was concordant, the diagnostic process was more straightforward, but not simplistic. And this is the first reported case of a primary yolk sac tumour in the abdominal wall of an adult.

INTRODUCTION

Yolk sac tumour (YST), also known as endodermal sinus tumour or primitive endodermal tumour, is a rare and highly malignant tumour of germ cell origin. It is common in infant and adolescent children with a median age at diagnosis of 19 years, with 40% of patients diagnosed in the prepubertal period [1,2]. This tumour occurs mostly in the ovary and the testis, and it rarely occurs outside the gonads, with only 10–15% of Yolk sac tumour (YST)cases reported locally and abroad [3]. Extragonadal YST can occur in the mediastinum, retroperitoneum, cervix, sacrococcygeal region, vagina, omentum, liver, and urachus [4-14]. Moreover, the clinical manifestations vary

according to the site of tumorigenesis, which often makes the clinical diagnosis difficult. Some cases lack typical structures for proper morphological observation, while others have diverse pathological morphology.

Yolk sac tumours arising in the abdominal wall are even rarer, with less than 2 cases of primary abdominal wall YST reported in the literature [15,16]. Here, we report a special case of primary YST in the abdominal wall of a 20-year-old woman, which to the best of our knowledge, is the first report of this disease in an adult.

CASE PRESENTATION

Chief complaints

A 20-year-old Chinese woman was admitted to the First Affiliated Hospital of Jinzhou Medical University in January 2022 with a lower abdominal mass, which had progressively enlarged since November 2021.It was not painful or itchy.

History of present illness

Palpation revealed that the mass was located in the hypogastric region, with a size of about 5×3 cm, The lesion was well defined, was not capsulated, normal skin without redness or swelling, poor range of motion, and no tenderness.

History of past illness

The patient has no past medical history.

Personal and family history

The patient denied any family history.Healthy parents, no siblings.

Physical examination

On physical examination, the vital signs were as follows: Body temperature, 36.3 ℃; blood pressure, 113/63 mmHg; heart rate, 86 beats per min; respiratory rate, 20 breaths

per min. Palpation revealed that the mass was located in the hypogastric region, with a size of about 5×3 cm, no redness and tenderness.

Laboratory examinations

The results such as routine hematological testing, blood sedimentation rate, and tumor-associated markers were normal.

Imaging examinations

Because the location of the tumor was relatively superficial, there was no imaging information at the beginning. The Positron Emission Tomography-Computed tomography (PET-CT) examination revealed no abnormalities in the abdominal organs and other regions, so this case was considered a primary tumour in the abdominal wall (Supplemental figure1).

FINAL DIAGNOSIS

A primary yolk sac tumour in the abdominal wall

TREATMENT

The patient underwent subcutaneous tumor resection. The mass was found to be located in the deep layer of subcutaneous fat, and the entire mass was completely dissected and sent for biopsy. We found the cut surface of the tumour was greyish-red and greyish-yellow in colour, soft to medium in quality, with visible necrosis in some areas (about 10%) (Supplemental picture 2). It was first considered as a possible soft tissue tumour or metastatic carcinoma.

After the tumour tissue was resected, it was fixed with 10% neutral-buffered formalin and then embedded in paraffin blocks. The tissue blocks were cut into 4-μm sections. Then, the hematoxylin and eosin-stained sections were used for histological evaluation. Histological findings from the resected tumor specimen showed classic micro vesicular, solid trabecular, papillary morphology and cystically area at low

power magnification (Figure 1). The surgical margins were negative, and it had visible necrosis, with no vascular invasion.

The morphology of the tumour tissues was diverse (Figure 2), and the main microscopic manifestations were as follows: 1) Characteristic Schiller-Duval bodies, which had central capillaries surrounded by a layer of cuboidal or low columnar embryonal epithelioid cells attached to the periphery. 2) Loose reticular structure, composed of a basophilic myxoid matrix, reticular microcysts, with or without a slit-like structure; the capsule wall was covered with flat, pleomorphic, or mesothelioid cells, which often had large and dark nuclei and cysts of varying sizes. 3) Papillary structure, with papillae composed of connective tissue axes and overlying epithelioid cells, with significant cellular and nuclear pleomorphism; the connective tissue had varying degrees of hyalinisation. 4) Solid structure, with an area composed of aggregates of small epithelioid polygonal cells with clear cytoplasm, large nuclei, and prominent nucleoli. 5) Mucoid structure. A few cords and adenoids composed of epithelioid cells arranged in myxoid stroma were observed. 6) Eosinophilic globules or hyaline bodies.

Immunohistochemistry were performed to establish a definitive diagnosis. The detection of the antibodies was accomplished using the streptavidin peroxidase method[17]. Immunostaining showed that broad-spectrum CK, GPC3 was strongly positive in almost all neoplastic cells. SALL4 was also strongly positive in the nuclei of the neoplastic cells (Figure 3), while CD117 and EMA were positive in a little range of neoplastic cells（Because the positive range was minimal (around 5%), we have reason to suspect that they were false positives）. The percentage of cells that stained positively for Ki67 was about 40% . The expression of AFP, Vimentin, CD31, CD34, D2-40, S100, Oct3/4, Desmin, SMA, PAX8, WT-1, PLAP, ER, PR, and hCG was negative in almost all neoplastic cells.Because it was shown in the literature that nearly all YST expressed AFP, we performed the second stain of AFP, and the result was also negative. In addition, we sent the samples to "Cancer Hospital, Chinese Academy of Medical Sciences" for counterstaining, AFP was still negative in expression.

OUTCOME AND FOLLOW-UP

The tumour was then resected without significant bleeding, and the patient recovered well without any signs of recurrence up to now. Remission was achieved after radical surgery, the patient was in good health. At the 3-,6-,and 9-month follow-up, the patient did not show local recurrence or distant metastasis, and the serum AFP was within normal levels. The patient will be followed up with clinical examinations and serum AFP level measurements every 3 mo for the first 2 years and every 6 months thereafter for the next 3 years.

DISCUSSION

Based on the clinical information, histological features, and immunohistochemical staining profile described above, the tumour was diagnosed as a primary yolk sac tumour in the abdominal wall. YST is a common tumour in the gonads, especially in the ovary and testis, and is rarely found in regions other than the gonads, according to a few documented reports. Extragonadal YST may have derived from the malposition of germ cells during embryogenesis. During embryonic development, germ cells migrate from the yolk sac *via* the midline dorsal mesentery to the genital ridge. During migration, some germ cells may remain anywhere along the migration pathway and develop into germ cell tumours [18]. These tumour locations include the liver and sacrococcygeal, gastric, retroperitoneal, and other regions [19-22]. Even fewer cases have been reported of YST occurrence in the abdominal wall. To date, only 2 cases of primary abdominal wall YST have been reported in the medical literature (Table 1) [15,16]. Both reports describe tumours in female infants, while our case occurred in a 20-year-old adult. And the 2 cases all expressed AFP, while in our case AFP was negative in expression.Ultimately, we believe that this was also a special feature of our case that is different from previous YST cases.

Previous reports, including this case, have summarised the features of the primary abdominal wall YST, which often occurs in younger children. Macroscopically,

these tumours are typically well-circumscribed, round, or spherical. The cut surface can appear greyish-white, greyish-red, or greyish-yellow. The histological pattern of this tumour is similar to the patterns found in the gonads. In this case, because the mass was located in the lower abdomen, the initial consideration of the clinicians was a sebaceous gland tumour, soft tissue tumour, or metastatic carcinoma. Thus, the patient in this case was not tested for AFP before the surgery. After resection, the patient's serum AFP level was 2.33 ng/mL (normal range: 0–8.04 ng/mL).

To make a correct diagnosis, the metastatic YST from the ovary should first be excluded through detailed clinical examination and evaluation. Moreover, the Positron Emission Tomography-Computed tomography (PET-CT) examination revealed no abnormalities in the abdominal organs and other regions, so this case was considered a primary tumour in the abdominal wall. Other differential diagnoses include embryonal carcinoma, dysgerminoma, clear cell carcinoma, teratoma, and other types of germ cell tumours. These different types of tumours can be distinguished by their respective histological features and immunohistochemical results. Embryonal carcinoma may show a solid structure, and the CK, SALL4, and glypican-3 expression can be measured using immunohistochemistry. However in embryonal carcinoma, the cells were more atypical, with large nuclei, prominent nucleoli, and no basement membrane-like material. The immunomarkers CD30 and OCT3/4 were positive in embryonal carcinoma, even though these were rarely observed in YST. Compared with YST tumour cells, dysgerminoma cells were more polygonal, the nucleoli were more obvious, and most were arranged in solid, cord-like, small tubular, and insular patterns. Meanwhile, the YST mostly showed cystic reticular structures with single solid structure. As for the immunohistochemical analysis, D2-40 and OCT4 were positive, and glypican-3 and CK were negative in dysgerminoma, while D2-40 and OCT4 were negative, and CK and glypican-3 were positive in YST. Clear cell carcinoma had a more regular tubular structure, often accompanied by papillary projections, cells overlying the tubules were cuboidal, clear cytoplasm, or boot stud-like cells, and lacked the typical perivascular structure of yolk sac tumors.

Immunohistochemically, clear cell carcinoma was diffusely strong positive for EMA and negative or partially positive for GPC3, while our results showed that very few areas of EMA are positive, while GPC3 was diffusely strong positive. Cystic mature teratomas were generally composed of three well-differentiated germ layers (endoderm, mesoderm, and ectoderm), and tumors were often cystic with a relatively intact capsule. Not consistent with the tumor we saw.

At present, there is no specific tumour marker for YST. The immunostaining of YST often displays the broad-spectrum CK, GPC3, and SALL4 positivity, which can be helpful for its diagnosis. YST has variable morphological features: microcystic/reticular, glandular, and solid structures are the more common patterns, while papillary and hepatoid structures are less common. Mixed histologic patterns were present in two-thirds of the cases, while Schiller-Duval bodies were seen in one-fifth of the cases [6]. As for the solid patterns, the neoplastic cells usually have mostly pale to clear abundant cytoplasm, with frequent intercellular basement membrane deposits, rare microcysts, nuclear pleomorphism, and hyaline globules. Thus, the present case showed a typical structure of YST.

CONCLUSION

Extragonadal Yst are usually a diagnostic challenge, because they are infrequent, but also because a thoughtful and detailed differential diagnostic process must be performed. In this case, since the tumor presented classic YST features, and immunohistochemistry was concordant, the diagnostic process was more straightforward, but not simplistic. And this is the first reported case of a primary yolk sac tumour in the abdominal wall of an adult. As for the prognosis of the disease is generally poor, the authors could follow up the patient closely.

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