

A Rare Case of Unilateral Krukenberg Tumor in a 24 Years Old Patient - A Case Report

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Citation: Beharee N, Zhujun S, Hua WJ (2019) A Rare Case of Unilateral Krukenberg Tumor in a 24 Years Old Patient - A Case Report. J Oncol Res Ther 6: 1081. DOI: 10.29011/2574-710X.001081

Received Date: 18 June, 2019; **Accepted Date:** 08 July, 2019; **Published Date:** 12 July, 2019

Abstract

Objective: A Krukenberg Tumor is a malignancy in the ovaries originating from a primary site and has been reported to be most commonly bilateral and occurs most commonly at an average age of 45 years old. However, in this article, we report a very rare case of unilateral Krukenberg Tumor, originating from the stomach in a 24 years old Chinese patient.

Case report: The patient, 24 years old, unmarried, virgin, attended hospital with initial complaints of 'abdominal fullness' and self palpated abdominal mass, noticed over the past few weeks. Based on the pre operative examinations, the patient was diagnosed as having a right ovarian mass (?Teratoma), and the possibility of a malignant tumor was not excluded. The patient underwent a laparoscopic surgery aiming at excision of the ovarian tumor. Specimens were sent to the histopathological laboratory for further investigation.

Conclusion: It is very difficult to establish the diagnosis of a Krukenberg Tumor at first hand, as it is not always easy to find the primary lesion. Thus, careful radiographic and endoscopic exploration of the digestive system is necessary to detect the primary tumor. Immunohistochemical evaluation is also useful for determining the primary site of the adenocarcinoma. The treatment of Krukenberg tumor includes resection of metastases and of the primary tumor, most commonly located in the gastrointestinal tract [1-4]. However, as occurred in our case, the primary lesion often remains undetected.

Keywords: Krukenberg Tumor; Metastasis; Ovarian Cancer; Stomach Cancer

Introduction

Krukenberg tumors are uncommon and account for less than 2% of all ovarian tumors [1,3]. The primary site for most (70%) cases of Krukenberg tumors is the stomach, followed by the colon, appendix, and breasts. Adenocarcinomas of the small bowel are uncommon, with an incidence of 0.23 cases per 100,000 patients and Krukenberg tumors arising from adenocarcinomas of the small bowel are even more unusual [1].

Krukenberg Tumor was first described by Friedrich Ernst Krukenberg in 1896. He described 5 cases of atypical ovarian tumor while working as a student in the laboratory of the German pathologist Felix Jacob Marchand. He presumed this was a new form of primary neoplasm [1,2]. As described by him, the term

Krukenberg tumor should be restricted to ovarian metastatic carcinomas that exhibit signet cells and diffuse stromal infiltration [1]. However, it has often been broadly applied to any metastases to the ovaries, irrespective of the site of origin [1]. The frequent association with ascites and its bilateralism were what led Friedrich Ernst Krukenberg to find an explanation for these clinic pathologic features [2]. During his research, he also noted the following findings: ovaries retained their general shape; they had a "Knobby" but smooth surface; were microscopically characterized by firm areas alternating with myxomatous zones; lymph nodes were prominently involved and the presence of cells containing mucin pushing the nucleus on one side (signet ring cells) [2].

Case Presentation

Patient Miss. L.Y, 24 years old, nulliparous, virgin, attended hospital with initial complaints of 'sensation of fullness' and self palpated abdominal mass noticed few weeks back. Micturition,

defecation, sleep pattern, feeding was normal. There was no remarkable weight loss. She denied any febrile episodes and had no other relevant complaints. No significant medical history or family history of bowel, breast, or gynecological cancer was noted. She is a non-smoker and doesn't consume alcohol.

Upon examination, vital parameters (Temperature, Pulse, Blood Pressure, Respiratory Rate) were within normal limits. Physical examination revealed a palpable right lower abdominal mass, hard, mobile, non-tender with a mixed texture. Gynecological examination was limited as the patient was virgin.

Rectal examination was carried out, showing a normal size, anteriorly located uterus. A right adnexal mass could be palpated measuring around 15X10 cm in size, mobile, non-tender and of a mixed texture. The left adnexa were unremarkable. The patient was admitted in ward and further investigations were carried out.

Ultrasound: Anteriorly located uterus measuring 53*40*31 mm in size, myometrium was unremarkable. An area of mixed echogenicity was noted anterior to the uterus, measuring 124*79 mm in size, with well-demarcated boundaries. CDFI: No obvious blood flow signal was found, indicating a pelvic mass,? Teratoma.

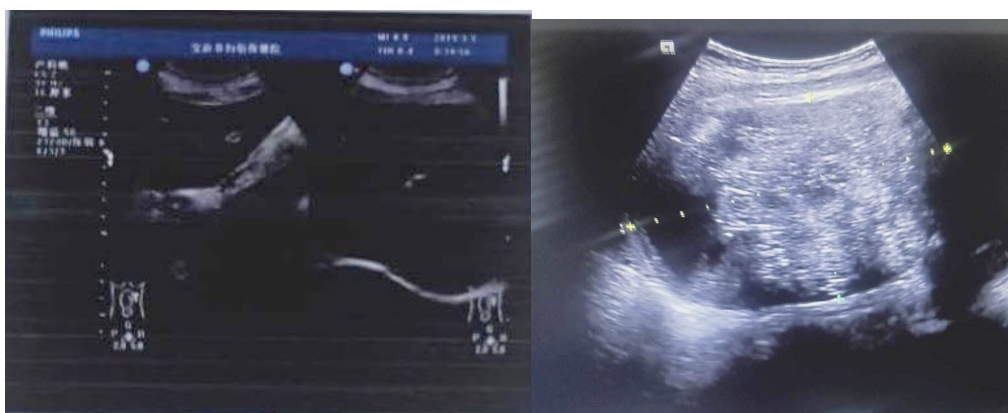


Figure 1: Ultrasound pictures.

Blood Investigations: Routine blood test, Urine analysis, Liver Function Test, Kidney Function Test, Fasting Blood Sugar were within normal limits. Tumor markers: CA125, HE4, ROMA, CEA, AFP, CA199 were normal.

ECG and Chest X-ray were normal.

Exploratory laparoscopic surgery was carried out. No obvious ascites was noted in pelvic and abdominal cavities. No obvious gross abnormalities were found in liver, stomach, spleen, omentum and intestine. The uterus was of normal size with normal appearance of left appendage and right fallopian tube. Right ovary visualized and a tumor of size 13*10 cm was noted, with smooth surface, greyish white in appearance and intact capsule. Abdominal cavity was irrigated with 200 ml saline and aspirate sent to the histopathological laboratory. No cancer cells were found in the aspirate.

Intra-operatively, in an aim to excise the tumor, the later burst accidentally. The content of the tumor were comparable to

'Rotten fish' like. The specimen comprising the tumor and all broken pieces were sent to the histopathological laboratory. The report came out as a Right Ovarian Adenocarcinoma. Based on the above finding, decision was made to convert to laparotomy for appendectomy and lymph nodes clearance. Para-aortic, pelvic, pre-sacral, abdominal, iliac lymph nodes clearance was carried out and multiple pelvic peritoneal biopsies were taken and sent to the histopathological laboratory.

Abdominal cavity drain was inserted and the surgery concluded. Post-operative progress was unremarkable with normal vital parameters and series of blood investigations were within normal range.

Postoperative histopathology report showed a right ovarian endometrioid adenocarcinoma grade 2-3. Tissues of the right ovary were arranged irregularly with maze-like cells. The atypia was obvious. No metastasis and no cancer cells were seen in the right fallopian tube, peritoneum, omentum and the lymph nodes.

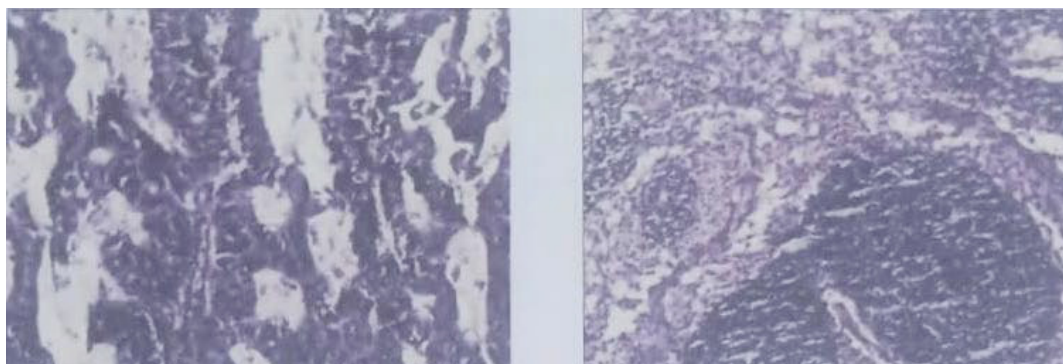


Figure 2: Post-operative Histopathology slides.

Immunohistochemical report: PMS2 (+); MLH1 (+); MSH2 (+); MSH6 (+); ER (-); CK20 (+); CDX-2 (+). The specimen provided showed a moderately differentiated adenocarcinoma of right ovary. Combining these findings, it indicates that the primary lesion could be in the intestine.

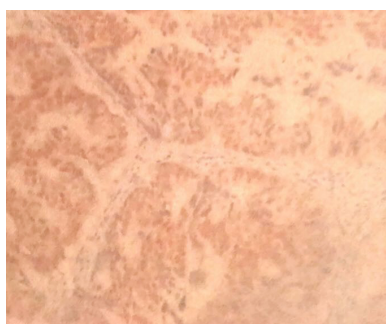


Figure 3: Immunohistochemical Slide Pictures.

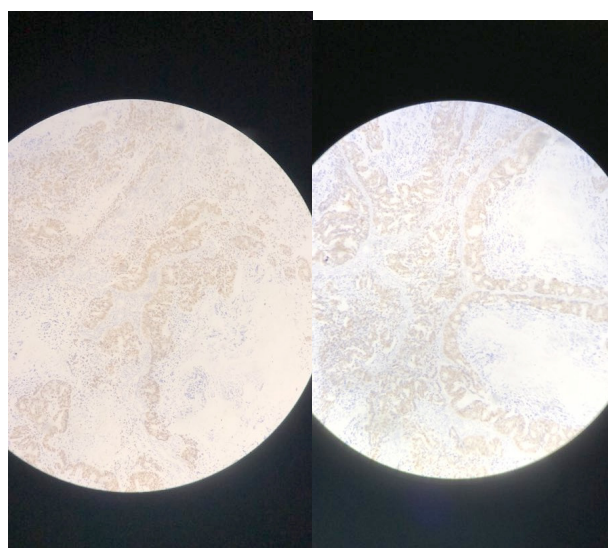


Figure 4: CDX 2.

The patient was discharged from the hospital and was transferred to another specialized hospital for further follow-up and management, whereby during further investigations, the primary lesion was found to be located in the stomach.

Discussion

Krukenberg tumors are rare (1-2% of ovarian tumors) and occur at an average age of 45 years old [1,3]. It is also more common in post menapausal women. However, in the above discussed case, the patient was 24 years old at the time of diagnosis and therefore represents one of the youngest case of Krukenberg tumor reported to date in the Jiangsu province, China. What make it a rare case, are the age and the unilateral lesion.

The diagnosis of a Krukenberg tumor can be established either preoperatively, intra-operatively or a few months post-operatively. Properly differentiating between primary and metastatic ovarian cancers is very challenging in many cases because of overlapping imaging results and sometimes the very small size of the primary tumor. However, masses that are bilateral, sharply delineated, purely solid, or predominantly solid lesions with necrosis favor the diagnosis of a metastatic ovarian tumor [1,5]. Therefore, the diagnosis of Krukenberg Tumor requires a very careful radiographic and endoscopic examination, alongside immune histochemical investigations to detect primary tumor [1]. In recent years (2009 - till date), despite being faced with all the difficulties in diagnosing, this hospital has been able to successfully diagnose 5 cases of Krukenberg Tumor from different primary lesions.

Diagnosis

Besides other criteria, the diagnosis of Krukenberg tumors depends to great proportion on the typical appearance of light microscopic features, such as densely fibroblastic and edematous stroma that appears diffusely infiltrated by malignant signet-ring cells arranged singly, in cords, or in nests [6]. Radiographic imaging, endoscopic examinations and immune histochemical

analysis are the other methods used in diagnosis.

Immunohistochemical staining was conceptualized and first implemented by Albert Coons in 1941. It is widely used in the diagnosis of abnormal cells such as those found in cancerous tumors. In immune histochemical analysis, particular cellular events have specific molecular markers that account for them [7]. Therefore, this test involves the process of selectively identifying antigens (proteins) in cells of a tissue section by exploiting the principle of antibodies binding specifically to antigens in biological tissues [7,8].

Immunohistochemical evaluation is also useful for determining the primary site of adenocarcinomas. For e.g. the immune phenotypes CK7 and CK20 and CDX2 are specific and sensitive for metastatic intestinal adenocarcinomas [9]. Specifically, the combination of CK7+, CK20-, CDX2- usually indicates an ovarian primary adenocarcinoma, whereas the combinations of CK7-/CK20+ and CK7+/CK20+ (particularly CK20+) strongly suggest a primary gastrointestinal adenocarcinoma [1,6].

Treatment

The treatment of Krukenberg tumor includes resection of metastases and of the primary tumor, most commonly located in the gastrointestinal tract [3]. However, the primary often remains undetected, as occurred in our case.

Chemotherapy is one of the lines of treatment, although its effectiveness is poorly established. The association of carboplatin and taxol can be used, depending on the primary origin [3]. The use of an association of drugs with a large spectrum of activity seems to be the right treatment choice for a metastatic tumor of unknown origin [3].

Prognosis

Currently, the prognosis of patients with Krukenberg tumors is extremely poor, with average survival time ranging between 3 and 10 months; Only 10% of such patients survive for more than 2years after diagnosis [10].

Conclusion

This case report presents a very rare case of unilateral right Krukenberg tumor, in a young patient, of reproductive age, which developed from a primary lesion in the stomach. Diagnosis in this case was challenging as it was a very atypical case.

It has been documented that radiologically such tumors present as bilateral solid ovarian masses (oval or kidney shaped). However, in this case, the ovarian tumor was unilateral (Right sided). The diagnosis was made based on the immune histochemical findings.

In conclusion, we must declare that the diagnosis of Krukenberg tumor is not always easy and obvious. To successfully diagnose this condition, a proper and careful radiographic and endoscopic exploration of the digestive system is required, so as to detect the primary lesion. Immunohistochemical analysis plays a crucial role in confirming the diagnosis and in determining the primary site of the adenocarcinoma.

Conflict of Interest Statement: The authors declare they have no conflicts of interest regarding this article.

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