ejpmr, 2020,7(7), 167-169

SJIF Impact Factor 6.222

EUROPEAN JOURNAL OF PHARMACEUTICAL AND MEDICAL RESEARCH

www.ejpmr.com

Case Study

ISSN 2394-3211

EJPMR

ECTOPIC PARATUBAL LOCATION OF ADRENAL TISSUE: CASE REPORT AND LITERATURE REVIEW

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Article Received on 02/05/2020 **Article Revised on 23/05/2020** **Article Accepted on 13/06/2020**

ABSTRACT

Aim: The aim of the study is to present a rare case of ectopic adrenal tissue localized adjacently to the right fallopian tube. **Material and methods:** Ectopic, paratubally located adrenal tissue was found accidentally during gynaecological surgery. Diagnostic laparotomy for a suspected disseminated abdominal tumor, with a primary origin in the internal reproductive organs, was performed on 3 February, 2020. **Results:** During the operation, 3,300 ml of fluid was evacuated from the peritoneal cavity. Considering the advanced stage of cancer, right appendages were removed and the parietal peritoneum sampled. Histopathological examination of the fragment adjacent to the right fallopian tube found ectopic adrenal tissue. **Conclusions:** Literature review found that in women the ectopic adrenal tissue in the internal reproductive organs isdetected accidentally. In the presented case the anomaly was also found by accident.

KEYWORDS: Ectopic adrenal tissue / Ectopic paratubal adrenal cell.

INTRODUCTION

The ectopic location of adrenal tissue is estimated at 1%in the adult population. In neonates, it occurs in about 50% cases. With time it atrophies and disappears in infancy. The anomaly is most frequent in boys.[1,2,3]

Ectopically located adrenal tissue in the internal reproductive organs is defined as very rare and is most frequently detected in the broad ligament.[4,5,6]

Embryogenesis of the adrenal cortex begins at about 4-5 weeks of foetal life arising from the mesoderm. An isolated clump of cells appears within the urogenital ridge, known as the adrenal-gonadal primordium. This tissue gives rise to the foetal adrenal cortex and to Lyedig cells. At 7 weeks of gestation, sympatho-adrenal cells migrate into the adrenal primordium. In later stages of embryonic development, the cortex engulfs, and ultimately encapsulates the entire medulla. Shortly after, toward the end of month 3, a second wave of cells from the coelomic epithelium (mesothelium) penetrates the mesenchyme and surrounds the original acidophilic cell mass. These smaller cells form the definitive cortex of the gland. At birth the structure of the external glomerular zone is still underdeveloped, but the zona fasciculata is seen readily and is directly continuous with the fetal zone. In the second month after birth the fetal zone begins to regress, the zona glomerulosa and fasciculata develop, zona reticularis develops, too.

During embryological development, medullary tissue penetrates into cortical tissue.

During this process, small fragments may separate. The disadvantages of this penetration process can lead to small separating fragments later migrating with the gonads.[7]

The development of genitourinary and adrenal organs is closely related. Therefore, ecotopic adrenal tissue can be found in the retroperitoneal space, mainly around the kidneys and adjacently to the adrenal glands.[4-8] Thus the ectopic adrenal tissue (mainly the adrenal cortex) can be found anywhere along the embryonic migration pathway.

Rare localization applies to other organs such as the renal parenchyma and distant organs such as stomach and liver.[9-14]

Abnormally located adrenal tissue is classified as ecotopic (additional) iflocated near the adrenal gland and along the gonads descending route. The location of this tissue in other organs was defined as the heterotopic. Referring to all such locations, Rosati J. uses the term ‘adrenal cortex heterotopia’.[15,16]

The histogenetic hypothesis of adrenal heterotopy assumes those changes originateat the primary adrenal stage and subsequently migrate to adjacent or distant organs.

Another hypothesis assumes the possibility of local formation of the heterotopic adrenal cortex from mesenchymal stem cells, among others from the placenta, differentiating into different mesodermal cell lines. This explains the cases of its occurrence in the placenta. The detected heterotopic adrenal tissue contains the adrenal cortex. Only in one case of paratracheal location it was found to contain the adrenal core tissue.[15]

It is believed that the occurrence of adrenal heterotopy is associated with many factors (multifactorial) caused by genetic and epigenetic changes.[17,18]

In general, heterotopic adrenal tissues are not related to any specific disease symptoms, are clinically insignificant, and are detected accidentally during surgery. In some cases, this tissue may be hormonally active and cause Cushing's syndrome.[19-21]

In few cases, the risk of cancer transformation was suggested.[21,22] and tumor transformation of ectopic adrenal tissue was reported by some authors.[5, 23]

Heterotopic localization of the adrenal glands is identified and confirmed by histopathological and immunohistochemical testing, like alpha inhibin immunoreactivity, Melan-A, Cam5.2, calretinin reactivity, HMB45, and CD10.[24]

MATERIAL AND METHODS

A 68-year-old woman was hospitalized in the Internal Medicine Department of the regional hospitalfrom 13 to 22 January, 2020. Prompt imaging and laboratory diagnostics was carried out, including chest X-ray, ultrasound of the abdomen, and CT of the chest, abdomen and pelvis with contrast.

Findings. Visible enhanced ascites and fluid in both pleural cavities (much more on the right) with atelectasis of the adjacent parenchyma. In the right lung, possible focal lesion within atelectasis. Enhanced ascites - fluid layer in the abdomen up to 95 mm.

Lab tests results.Ca 125 - 294.4 U / ml (normal value - <1.20 U / ml).

Protein electrophoresis showed a reduced albumin / globulins ratio of 53.1% (norm 60.3 - 72.8%). Increased level of globulins:alpha -1 globulin 3.4% (norm 1.0-2.6%), beta globulin 2 - 6.4% (norm 2.2-5.7%), and gamma globulin -16.9 % (norm 6.2 - 15.4%). The patient was discharged with the diagnosis of ascites and suspected cancer of the reproductive organs. She was referred for further diagnosis and oncological treatment to the Department of Gynecology and Obstetrics.

CASE REPORT

A 68-year-old patient was admitted to the Ward on 24 Jan., 2020 with a history of twonatural deliveries and one spontaneous miscarriage. After admission, the diagnostics was extended: gastroscopy, colonoscopy, histopathological examination of fluid obtained during peritoneal puncture was performed. Colonoscopy reaching the splenic fold did not reveal rectal or colorectal lesions. Further on, an acute, non-insufflectable bend was found which prevented the rest of the intestine from being examined. Gastroscopic examination did not reveal any lesions in the stomach and duodenum. The examination of peritoneal fluid found no anomalies.

The patient underwent surgery on 3 Feb., 2020. The abdominal cavity was opened with a lower median incision outreaching the navel. About 3,300 ml of transparent straw-colored fluid was evacuated, and taken for histopathological examination. Cancerous infiltrates on the walls of the small and large intestine were found. The greater omentum was shrunk, infiltrated, infiltrates involving the organs of the upper half of the abdomen. The sigmoid infiltrated reaching the left appendages. Small bowel infiltration involving the bottom of the uterus and the parietal peritoneal wall covering the bladder. The uterine body was not enlarged, of limited mobility. Due to cancer advancement, the surgery was limited to the excision of the right appendages. Parietal peritoneum was biopsied. A safety drain was inserted into the peritoneal cavity. The abdomen was closed in layers. The skin was sewn with single stitches.

The postoperative course and wound healing was uneventful. The patient was discharged from the Ward on the seventh day after the surgery.

Histopathological findings

Peritoneal fluid - *Probabiliter cellulae carcinomatosae solitariae.*

Right ovary - In the cortical layer of the ovary, a small focus of serous cancer extending beyond the capsule. Foci of cancer visible in the fibrous-fatty tissue around the ovary and around the fallopian tube. Angioinvasion.

Tubal and tissue fragment –single focus of adrenal tissue.

Fragments of the parietal peritoneum from the left and right side - Cancer infiltration in the fibrous-fatty tissue of the parietal peritoneum.

Diagnosis. The picture suggested primary right ovarian cancer, metastasis from theleft ovary, or primary peritoneal tumor.

Oncological pharmacological treatment was not implemented as the patient died on 10 March, 2020.

DISCUSSION

The occurrence of ectopic paratubal location of adrenal tissue is described as extremely rare. Moreover, preoperative diagnosis poses numerous difficulties.[25]

In the literature, the coexistence of ectopic adrenal tissue with malignancy or borderline malignancy affecting the reproductive organs is also described as rare.[22]

Similarly to literature reports, adrenal heterotopy in the area of the right fallopian tubewas accidentally detected.[24]

The advanced stage of underlying disease produced unsuccessful outcome of the implemented treatment. We report on the case due to the rarity of adrenal tissue ectopy at paratubal location and its coexistence with ovarian malignancy.

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