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Case Report

Benign cystic teratoma of fallopian tube with shadow cell differentiation – a rare diagnostic entity with review of literature

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ABSTRACT

A 26 year old gravida 2 para 1 presented to obstetrics department at full term with an unremarkable USG. LSCS was performed for CPD. A fimbrial cyst was discovered intra operatively and excised and sent to us for histopathological examination which showed derivatives of all 3 germ cell layers with presence of shadow cells surrounded by basaloid cells and giant cell reaction. Objective was to report a case of mature cystic teratoma of Fallopian tube with shadow cell differentiation.

Keywords: Fallopian Tube, Shadow cell differentiation, Teratoma

INTRODUCTION

Neoplasms of uterine tube, both benign and malignant, are the least common tumours of the female genital tract.[1] Tubal teratoma is quite rare and only 54 cases have been reported in literature till date. Generally they are benign and found incidentally.[2]

Mature cystic teratoma, also known as dermoid cyst, originate from primordial cells and are composed of well differentiated derivatives of any combination of three germ layers i.e. ectoderm, mesoderm and endoderm.[3] Shadow cell differentiation has been described in testicular dermoid cyst but never in Fallopian tube teratoma. So called 'shadow cells' are distinctive necrotic keratinized cells having preserved cell shape, small empty spaces left after disappearance of the nuclei with eosinophilic cytoplasm.[4] Here we present a case of incidentally discovered benign cystic teratoma with shadow cell differentiation of unilateral Fallopian tube in a term pregnant female.

CASE REPORT

A 26 year old female G_2 P_{1+0} with unremarkable menstrual history underwent lower segment caesarean section for cephalo-pelvic disproportion and pregnancy induced hypertension at Burdwan Medical College and Hospital.

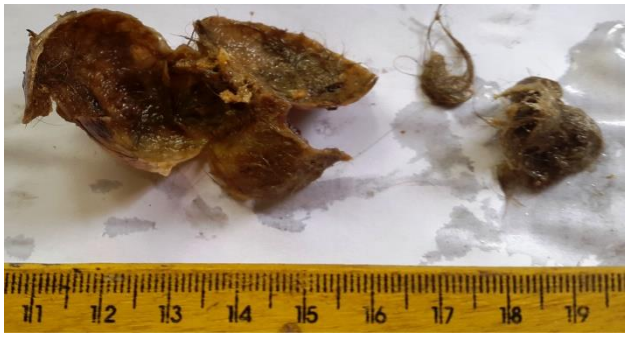

Figure 1: Unilocular cyst containing hair and teeth structures on gross.

Intraoperatively a freely mobile fimbrial cyst was discovered which was attached to the wall of left adnexa. Grossly, the brown coloured cyst measuring 7cm × 5cm in diameter, having 10 ml yellowish turbid cheesy material in the unilocular cyst cavity, was found to contain hair and teeth on cut section (Figure 1).

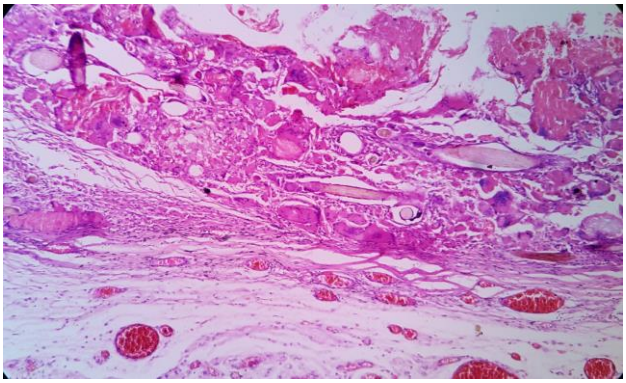

Figure 2: Fibrous cyst wall containing keratin lamellae with prominent foreign body type of giant cell reaction [Hematoxylin & Eosin 100X].

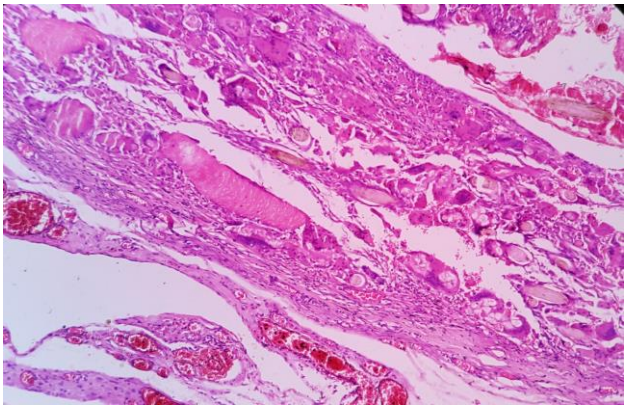

Figure 3: Shadow cell differentiation within the keratin lamellae in the cyst wall along with hair structure and giant cell reaction [Hematoxylin & Eosin, 100X].

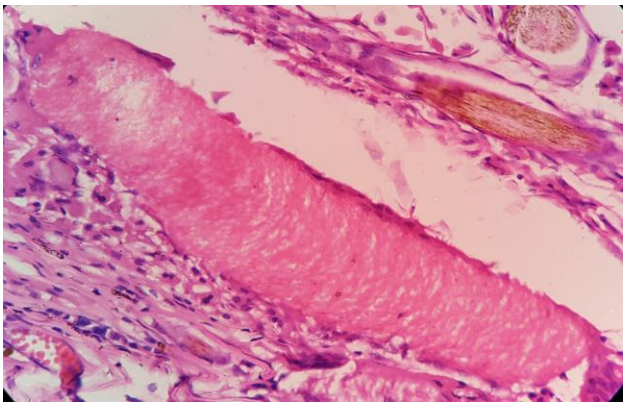

Figure 4: Shadow cells among keratin lamellae, partially surrounded by few basaloid cells [Hematoxylin & Eosin, 400X].

Histopathologic examination showed that the cyst wall consisted of fibrous tissue, skin adnexal structures, cartilage and focal group of shadow cells among keratin lamellae, surrounded by basaloid cells with a prominent giant cell reaction (Figure 2-4).

Final diagnosis of a Benign Cystic Teratoma of Fallopian tube with Shadow Cell Differentiation was made. Transvaginal sonogram, done on 40th day post-surgery, revealed an unremarkable uterus, adnexae and POD.

DISCUSSION

The first case of benign teratoma of fallopian tube was reported by Eden and Lockeyer in 1865.[5] Since then a large number of cases have been reported. The first 25 cases were collected by Aaron in 1941.[6] Mozzarella et al reviewed the literature and gave a detailed account of both solid as well as cystic teratomas.[1] However, in 1964, Legerlotz pointed out that some of the tumours might be cystic teratomas of the ovary with extention into the uterine tube.[7] Till date 54 cystic teratomas of the fallopian tube have been reported.[2]

The age of the patients with tubal teratoma range from 21 to 60 years.[1] Tubal teratomas are rarely diagnosed pre-operatively. The most common symptoms are pelvic or abdominal pain, dysmenorrhoea, menstrual irregularity, leukorrhoea and post-menopausal bleeding. But none of the symptoms are specific and diagnostic.[1] Teratomas are a heterogeneous group of germ cell tumors which predominantly arise in the gonads. However they can arise in any age and at any extragonadal midline site.[8] On macroscopic examination teratomas may be cystic, predominantly solid or a mixture of solid and cystic with variable amount of necrosis.[8] On microscopic examination, both mature and immature teratomas contain derivatives of all 3 germ layers. Therefore different epithelial, mesenchymal and neural tissue components may be present.

Mature teratomas consist of haphazardly arranged, heterogeneous but completely differentiated somatic tissues derived from all three germ cell layers, which may be foreign to the specific tumour site. The disorganized pattern of arrangement of the various components has been termed ‘histological potpourri’. Complex structures like intestinal segments and pancreatic tissue with functional islets of langerhans have also been reported in some tumours. Large dermoid cysts are frequently lined by pseudostratified squamous epithelium with hair and other cutaneous adnexal structures like sweat and sebaceous glands.[8]

Immature teratoma on the other hand contains atleast one foci of immature or embryonal or incompletely differentiated tissue component.[9-12] Most commonly the immature component is primitive neuroectodermal tissue which can present as primitive neurotubuli, rosettes or neuroblastomatous foci. The degree of immaturity

correlates with mitotic index. The mitotic activity or the rate of proliferation can be studied by immunostaining to assess the Ki67 index.[8]

Malignant elements such as seminoma, yolk sac tumour, embryonal carcinoma or a varying combination of any of these may be present in these groups of neoplasms.[8] The heterogeneous clinical and histological pattern correlates with the complex histogenesis of germ cell tumors which develop from the totipotent germ cells.[13] Thus, teratomas always reflect variable degrees of tumor progression and regression as well as differentiation and de-differentiation.

Shadow cell differentiation has been traditional diagnostic clue for pilomatrixoma, matricial carcinoma and other cutaneous tumours with differentiation towards hair matrix. It is also seen in extra cutaneous lesions like craniopharyngeoma, odontogenic lesions, testicular dermoid cyst, ovarian adenosquamous carcinoma, pilomatrixoma, uterine lesions (adenosquamous carcinoma, carcinosarcoma, atypical hyperplasia), prostatic squamous cell carcinoma, urothelial carcinoma of urinary bladder with squamous metaplasia, adenosquamous carcinoma of colon, small cell carcinoma of gall bladder with squamous metaplasia and dermoid cyst of brain.[4] SCD has also been reported in a case of pancreatoblastoma by Klimstra et al.[14]

The presence of shadow cells is considered to be an indicator of hair matrix differentiation in cutaneous tumours.[15,16] However in extracutaneous lesions, shadow cells when found, are generally not associated with other structures of hair matrix like small dark matricial cells, supra matricial cells, melanocytes or well differentiated pilosebaceous units. So in extra cutaneous tumours, Shadow Cell Differentiation as an indicator of hair matrix differentiation, does not seem a valid proposition. However in such lesions, shadow cells have often been found in squamous cell areas that show lamellar keratinisation (as in squamous morules of adenoacanthoma).

In the absence of other signs of hair differentiation, the shadow cells in these tumours probably represent a mode of karatinization with no histogenetic relation to hair matrix differentiation. However in their case report, Zamecknik et al presented a teratoma which contained shadow cells along with skin structures (similar to this case) and proposed that these findings should be interpreted in analogy with cutaneous lesions i.e. they probably indicate hair matrix differentiation.[4] The absence of close association between shadow cells and viable epithelial cells could be attributed to limited sampling. Hitchcock et al reported presence of shadow cells inside hair follicles in dermoid cyst of the brain.[17] This observation indicates that the close relationship between shadow cells and hair follicles, as is known in skin lesions, also applies to well differentiated extra cutaneous lesions like mature cystic teratoma.

Most of the tubal teratomas are found incidentally during pelvic operations and mostly they are thought to be ovarian neoplasm rather than primary fallopian tube lesion.[7] Although rare, the possibility of fallopian tube teratoma should be considered if a tubal mass is found. In 1998, Hseih et al reported a case of benign cystic teratoma of unilateral fallopian tube in a term uterine pregnancy.[18] Shadow cell differentiation although not commonly found in a teratoma, does indicate differentiation towards hair matrix. So, consequently in absence of hair or pilosebaceous unit in a teratoma due to inadequate sampling, e.g. if only nests of basaloid cells with shadow cells are found, then it should be highly indicative of an attempt at differentiation towards hair matrix which is ectodermal in histogenesis.

CONCLUSION

Tubal Teratomas are rare entities which are rarely diagnosed pre operatively. Though our case was an incidental finding, yet it can be considered as a rarer histomorphological pattern in a rare tumour as heterogeneous histomorphology of teratomas will provide the biologic basis for rational clinical diagnosis, treatment strategies as well as its molecular biological behavior in future days.

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