



## Mature cystic teratoma with synchronous immature teratoma of opposite ovary in a nine year old girl – a varied presentation of common tumor – Case report with review of literature

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### ABSTRACT

Teratomas are most common germ cell tumors with two or more germ cell layers. Mature teratomas are common compared to Immature teratomas. These tumors are common in young and adolescent girls and induce large impact on their reproductive life and on the natural life span, which in turn depends upon the complications in

case of Mature teratoma; Grading and staging of tumor in Immature teratoma. We present here a case of Mature cystic teratoma in one ovary and Immature teratoma in opposite ovary in a nine year old girl. A common tumor with uncommon and varied presentation.

**Key words:** *Benign tumor, Immature teratoma, Malignant tumor, Mature teratoma, Young female*

### Introduction

Teratomas are composed of recognizable tissues of ectodermal, mesodermal and endodermal origin, in any combination. They are common, usually benign and are inert but rarely produce remarkably bizarre and varied syndromes, reflecting the diverse potentials of germ cells[1]. Immature teratomas are common germ cell tumors comprising of two or more germ cell layers (ecto, meso or endoderm) derived from a pluripotent malignant precursor cell[2]. Mature teratomas account for approximately 15% of all ovarian tumors[3], whereas immature tumor is rare with less than 1% of ovarian tumors[2]. Mature teratomas are classified as cystic, solid or monodermal. Immature teratomas show only solid mass[4]. In mature teratomas – most commonly mature ectodermal elements like skin, hair, sweat and sebaceous glands are notified, whereas in Immature teratomas – tissues with partial somatic differentiation identical to that of foetal tissues are found[5]. Majority of teratomas are benign and about 43-70% are found and diagnosed in reproductive years. Fifteen percent of cases develop bilaterality. Highest frequency is notified during adolescence[6]. Immature teratomas account for about 10-20% of all ovarian neoplasms in women of less than 20 years age. The peak incidence is observed between 15-19 years. Thirty percent of deaths are reported from this ovarian neoplasm in this age group[7]. Of all mature cystic teratomas 1-3% show malignancy or

transformation. This transformation ratio also decreases in tumors seen in young patients[8]. Mature cystic teratoma may present as mass per abdomen, pain abdomen and compression syndrome depending upon its size and site of obstruction[1]. Immature teratoma may present as calcified pelvic mass, abnormal uterine bleeding or pelvic pain[9]. Laparotomy is the treatment of choice for management of mature teratoma – dermoid cyst as there exists risk of chemical peritonitis due to spillage of contents. Thus, conservative surgical excision with salpingo-oophorectomy is performed in majority of cases[10]. In case of immature teratoma – foundations for treatment have been steadfast throughout decades: conservative primary surgery usually involving unilateral (affected) salpingo-oophorectomy followed by combination chemotherapy is practiced[2]. We present here, a case of bilateral teratoma with mature cystic teratoma (dermoid cyst) in one ovary and immature teratoma in the contralateral ovary in a nine year old girl.

### Case report

A nine-year-old girl presented with pain in lower abdomen for the previous 3 months. The pain was of variable intensity – dull to colicky in nature, aching and non-radiating type. The patient has not yet attained menarche. No significant medical or surgical history was notified.

Physical examination revealed soft abdomen, without any rebound tenderness or guarding. There was an abdominopelvic mass – 6 cms. below right costal margin. The mass was mobile and of firm, solid to cystic in consistency. The speculum examination showed no vaginal discharge. Vaginal examination revealed a large right adnexal mass (6.0 x 5.0 cms) and a small adnexal swelling over left side (2.5 x 2.0 cms).

Ultrasound abdomen showed a right ovarian cyst 6.5 x 5.5 cms. with solid and cystic areas. Left ovary was slightly enlarged 3.0 x 2.8 cms with pure solid component. Uterus was normal. A provisional diagnosis of bilateral ovarian tumor(? dermoid cyst) was made and patient was subjected for further routine investigations. AFP (Alpha fetoprotein) and CA-125 was normal and all pre-operative routine investigations were in the normal reference range.

The patient underwent laparotomy. There was no free fluid in abdomen. A complex cyst was resected along with right ovary measuring 6.8 x 6.0 cms. Thus, right salpingo-oophorectomy was done. Consequently the left ovary was exposed which revealed a small almond shaped solid mass of 3.5x2.8 cms. Left salpingo-oophorectomy was also performed, followed by omentectomy and periaortic bilateral lymphadenectomy. All the excised tissues were subjected for histopathological examination.

The resected tissue specimen was received as four tissue masses separately.

*Gross examination revealed: -*

1. The cystic mass measuring 7.0 x 6.0 x 4.5 cms with smooth external covering capsule showing enlarged blood vessels and attached fallopian tube (swollen and oedematous) measuring 4.0 cms in length. On cut section of cystic mass there are complex solid and multiloculated cystic areas with large aggregates of hair and pultaceous material. The solid hard areas showed glistening knob like cartilaginous areas - "umbo" admixed with fleshy necrotic and hemorrhagic areas (Figures: 1 & 2)
2. The second sample was solid small grey white mass of 3.5 x 2.8 x 2.0 cms, showing smooth external surface. Cut section showed heterogenous grey white to grey yellow areas. The attached fallopian tube is 1.0 cm. in length and normal. (Figure: 3)
3. The third sample was omentum - 10 x 3 cm 1.2 cms – grey white in colour
4. The fourth sample was an irregular mesenteric mass 8.0 x 7.0 x 2.0 cms – grey white in colour. Cut section showed four grey white solid lymph nodes.

*Microscopic Examination showed: -*

1. Right ovary (large mass) – showed cyst wall lined by stratified squamous epithelium with underlying sebaceous glands and hair shafts. Focal areas revealed cartilaginous and bony elements, thyroid tissue and neural tissue. There are at places pools of desquamated keratin and secretions from sebaceous glands along with hair filling the contents of cyst. (Figure: 4)
2. Left ovary (small mass) - showed cyst wall lined by hyperplastic stratified squamous epithelium showing atypical epithelial cells with focal areas of hemorrhage and necrosis surrounding small islands of neuroectodermal tissue in the form of rosettes and tubules and cellular foci of glial cells with numerous mitoses. Focal areas also showed islands of immature bone, cartilage and glandular structures distributed through a poorly differentiated fibrous connective tissue stroma. (Figure: 5)
3. Omentum showed mature fibrofatty tissue and was free from any other elements. No infiltration was observed.
4. All the four lymph nodes resected showed only reactive hyperplasia. No infiltration or secondary deposits were observed.

The patient was free of any post-operative complications and was discharged after 5 days. The recovery was uneventful as there was no discharge or post-operative pain during removal of sutures on 10<sup>th</sup> post-operative day.

With all the above investigations performed the patient was finally diagnosed as individual with left immature solid teratoma essentially malignant grade-I with right mature cystic teratoma – dermoid cyst and was referred to clinical oncology for adjuvant chemotherapy and has received six cycles of treatment with vincristine / actinomycin / cyclophosphamide. At present, the patient is being called for follow-up every six months and is without any evidence of recurrence or residual disease.

**Discussion**

The term teratoma was derived from greek root "teratos" which means "monster"[11]. Ovarian mature teratoma is common germ cell neoplasm accounting for 15-45% of all ovarian tumors in contrast to immature teratoma which accounts for only 1% of all ovarian tumors. It is as such common to find an immature teratoma in one ovary with mature teratoma in the contralateral ovary[12]. But, such cases are sparsely found in the literature.



Figure: 1. Excised right ovary with smooth glistening surface and superficial vessels.



Figure: 2. Cut section of right ovary showing numerous cysts and solid areas. Cysts filled with pultaceous material and hair & glistening cartilaginous areas.

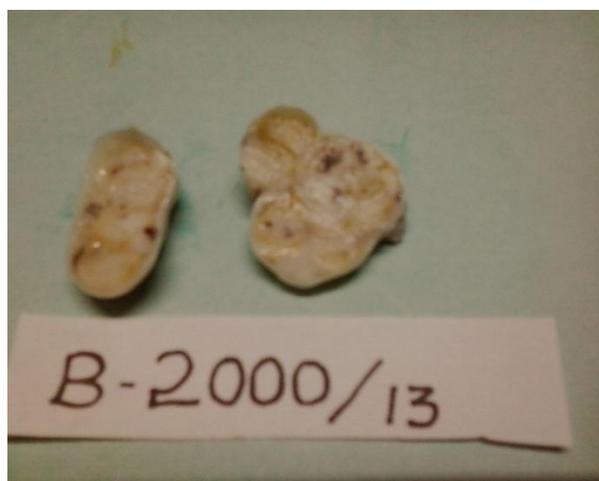


Figure: 3. Excised left ovary- almond shaped, heterogenous, grey white, solid areas

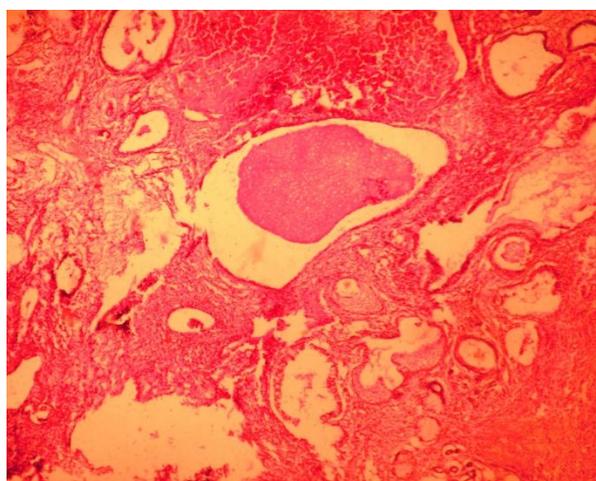


Figure: 4. Microphotograph showing bronchial epithelium, glands, colloid and mucin and sebaceous glands – mature cystic teratoma. (H&E, X100)

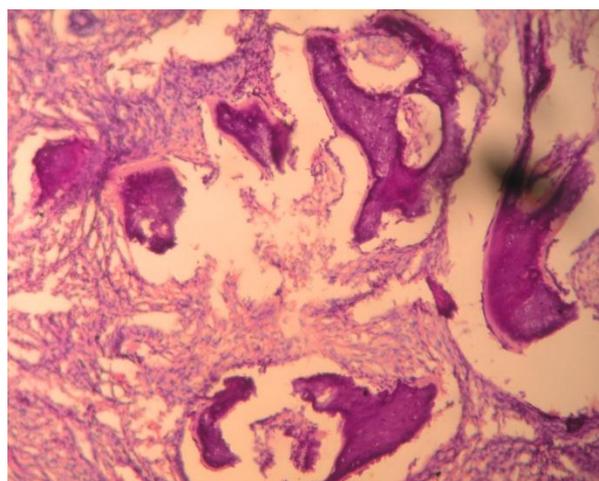


Figure: 5. Microphotograph showing mature cartilage, bony spicules & fibrous stroma studded with hair follicles. (H&E, X100)

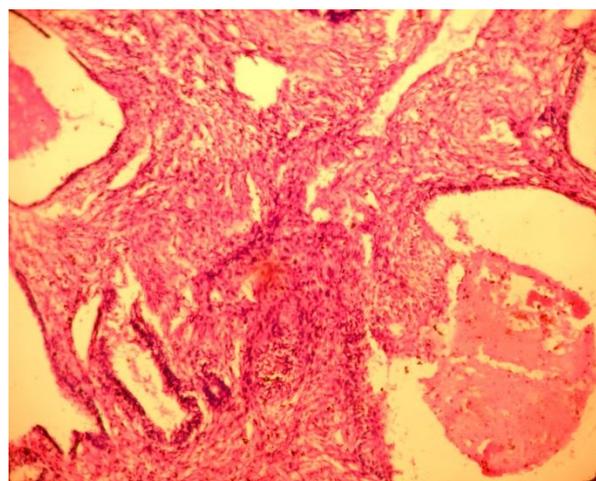


Figure: 6. Microphotograph showing immature neuroepithelium and a few rosettes surrounded by fibrous connective tissue. (H&E, X100)

Mature cystic teratomas are mostly diagnosed incidentally during physical examination, radiological evaluation or during abdomino-pelvic surgeries for other ailments. In literature asymptomatic tumors are found to be present in about 6-64.5%[13,14]. Immature teratomas mostly present with abdominal pain as chief complaint followed by abdominal mass. Radiological evaluation suggests unilateral immature teratoma as common lesion with contralateral dermoid cyst in 7.1% of cases as is reported in the present report. Similar case series was submitted by Norris et al[15] and Wisniewski et al[16].

Mature teratoma presents with unique features which demands appropriate diagnosis through ultrasound examination and other improvised radiological investigations. Ultrasound examination of immature teratoma appears similar to mature teratoma. Thus, CT and MRI with contrast imaging help to find a way for better distinction [17].

Germ cell tumors are serologically evaluated by tumor markers. In case of immature teratoma AFP is widely used. It has been suggested that the AFP level in immature teratoma is not correlated to either stage or grade of the tumor. Thus, AFP plays a limited role in evaluation and management of germ cell tumors[18-21].

Even though the laparoscopic approach is generally preferred for mature cystic teratoma – the rupture of cysts are more frequently reported in laparoscopy than in laparotomy. Thus, the standards of the oncologic surgery must be applied to prevent spillage[15]. Immature teratoma is treated based on FIGO (International Federation of

Gynecologists & Obstetricians) staging and grading of tumor (Table: 1) as it occurs in young patients. Preservation of fertility is an important factor in management[22].

Grade I & FIGO Stage I – unilateral oophorectomy [23]

Grade II / III with advanced stage – adjuvant chemotherapy in addition to surgery (bleomycin, etoposide and cisplatin) [24]

The current combination of chemotherapy results in overall disease free survival rate of >95%[25].

However, at the outset, the final diagnosis is confirmed by histopathological examination of excised specimen showing mature contents of all epithelial elements in mature teratoma and immature / fetal tissue elements in immature teratoma. The amount of neuroepithelium correlates with survival and is the basis for grading these tumors. In turn, the only type of neural tissue that is to be evaluated in grading the immaturity of tumor is primitive neural tubes and immature rosettes[1,15,26,27].

In mature teratoma, recurrence rate range from 3-4%[3]. Recurrence is seen more commonly in patients who are younger at the time of first diagnosis. The bilaterality and multilocularity of tumor also is an additive factor in recurrent cases[28]. Recurrence in immature teratoma is based on grade of tumor. Grade III tumors show high rate of recurrence [29]. Diagnosis of recurrence of immature teratoma by tumor markers appears to be more sensitive when combined with detection of CA-125, CA-153 & AFP[30].

**Table: 1. Grading of immature teratoma[1]**

Sl.no	Grade	Histopathological features	Prognosis
1.	I	Tumors with rare foci of immature neural tissue occupying less than 1 low power field / slide	Good
2.	II	Tumors with moderate quantity of immature neural tissue filling more than one but three or fewer low power fields / slide	Worse
3.	III	Tumors with large amounts of immature neural tissue occupy 4 or more low power fields / slide	Poor

## Conclusion

Ovarian teratomas commonly have a very indolent course and in majority of patients the diagnosis is made accidentally or incidentally. In this case report there are two different tumors showing similar origin with benign and malignant counter parts in opposite ovaries making it more challenging for evaluation and management. This case reflects the importance of early diagnosis in case of pelvic masses in children and adolescents which in turn helps to formulate appropriate management protocol causing least possible impact on life span and reproductive future in the young women.

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