

Adenocarcinoma (Somatic-Type Malignancy) in Mature Teratoma of Anterior Mediastinum

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ABSTRACT

Mature teratoma is a common anterior mediastinal tumour. However, occurrence of transformed malignant component within it is very rare. We report a case of a 32-year-old female presenting with dry cough and chest pain. Contrast-enhanced computed tomography (CT) showed a large thin-walled cystic lesion measuring 11.4cmx10.6cmx10.0cm in the anterior mediastinum. Right postero-lateral thoracotomy was performed and the tumour was completely excised. Histopathological examination of the excised specimen was suggestive of mature teratoma with transformed malignant component, adenocarcinoma (somatic-type malignancy). [*Indian J Chest Dis Allied Sci* 2013;55:39-41]

Key words: Adenocarcinoma, Mature teratoma, Mediastinum.

INTRODUCTION

In adults, the anterior mediastinum is the most frequent extragonadal site of origin of non-seminomatous germ cell tumours, representing 10% to 15% of all mediastinal tumours. Of those, mature teratoma is the histopathologic subtype, that is seen in 50% to 60% of patients.¹ Teratomas that undergo malignant transformation are rare, and comprise only 6% of all (gonadal as well as extra-gonadal) teratomas. Of the transformed teratomas, only 5% are of the mature type and the rest are of immature type.¹ It mostly arises from a pre-existing teratoma following chemotherapy and/or radiotherapy for patients with an initial malignant germ cell tumour.^{2,3} Naturally occurring teratomas that undergo malignant transformation in the mediastinum are extremely rare. We report a rare case of naturally occurring adenocarcinoma arising in an anterior mediastinal mature cystic teratoma with a review of the literature.

CASE REPORT

A 32-year-old female presented with occasional dry cough since four years and chest pain on the right side since one year. General physical examination was normal. Respiratory system examination revealed reduced air entry in the lower right-side of the chest with a dull percussion note; rest of the respiratory system examination was normal. Spirometry was

suggestive of moderately severe restrictive pattern. Chest radiograph showed homogeneous ground-glass opacity in the right lower lobe. Contrast enhanced computed tomography (CT) showed a large thin-walled cystic lesion of size 11.4cmx10.6cmx10.0cm in the anterior mediastinum, with a few thin septations, mildly compressing the right atrium and compressive atelectasis of the adjacent right lower lobe, suggestive of benign cystic lesion (Figures 1A and 1B).

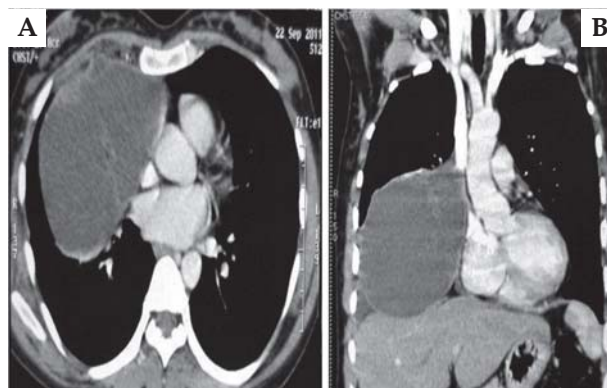


Figure 1. Axial (A) and coronal (B) multislice-CT reformatted post contrast images showing a well-defined, homogeneous, cystic, thin-walled, non-enhancing focal lesion with thin enhancing internal septations, in the anterior mediastinum.

Hydatid cyst and mediastinal thymic cyst were considered as differential diagnosis. Right postero-lateral thoracotomy was performed. The tumour, which was located in the anterior mediastinum,

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reaching upto the diaphragmatic surface, not invading the right lung and pericardial sac, was completely excised. Approximately 1.2 litre of sero-sanguinous fluid was drained out from the cystic mass. The excised specimen was sent for histopathological examination.

On gross examination, the cystic mass with drained out contents measured 12cmx8cmx5cm (Figure 2). No definite capsule or lung parenchyma were identified on the external surface. Areas of congestion were evident. The cyst wall was smooth measuring 0.2cm to 1cm in thickness with few yellow areas. No solid areas or papillary excrescences were noted on inner surface except for a 9cmx2cm ridge like elevation.

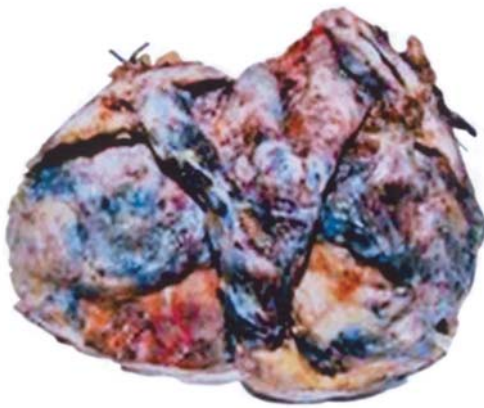


Figure 2. Gross specimen photograph showing large mass with congested external surface.

Microscopically, most of the mass was composed of fibrocollagenous tissue with congested vessels and chronic inflammatory cells. In sections obtained from the ridge like elevation that was evident on gross examination, stratified squamous epithelium, cystic spaces lined by tall columnar ciliated epithelium and

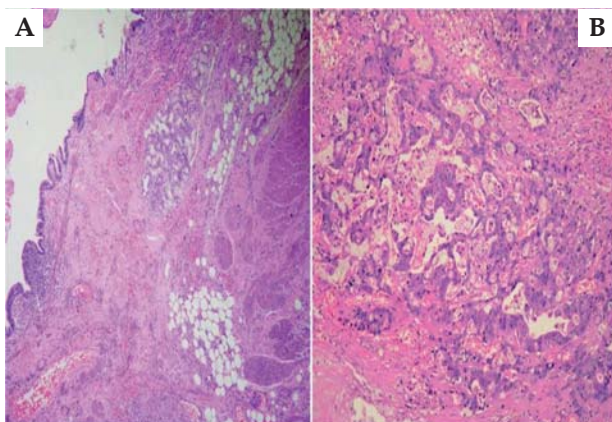


Figure 3. Photomicrograph showing fibrocollagenous tissue, adipose tissue and muscle bundles; focally lined by cuboidal to ciliated columnar epithelium (A) and areas of adenocarcinoma; (B) (Haematoxylin and Eosin x100).

focal mucin secreting epithelium, muscle bundles, adipose tissue and sebaceous glands were seen. Areas showing atypical cells in groups and glandular formations, suggesting adenocarcinoma were present in all the sections (Figures 3A and 3B). No bone or cartilage tissues were observed and there were no immature teratomatous elements or other germ cell components. The final histopathologic diagnosis was cystic teratoma with somatic-type malignancy (adenocarcinoma).

The patient had an uneventful postoperative course. She has not received adjuvant chemotherapy or radiotherapy. There has been no recurrence of the lesion and the patient is doing well on follow-up two months after the surgery.

DISCUSSION

Mature teratoma consists of fully differentiated adult tissues derived from more than one of the three embryonic germ cell layers (ectoderm, mesoderm, endoderm). It occurs in the gonads and also extra-gonadally in the midline, in the cranial cavity, mediastinum, and retroperitoneum.⁴ Pathologically, malignant teratoma can be divided into three types: immature teratoma; teratoma with other malignant germ cell tumour components such as yolk sac tumour, embryonal carcinoma, choriocarcinoma, seminoma; and teratoma with malignant transformation.³ Non-germ cell malignancies arising from a pre-existing teratoma are at present described as teratoma with somatic-type malignancies by the current WHO classification.⁴ It is postulated that somatic-type malignancy develop from either malignant transformation of pre-existing teratomatous elements, or by differentiation of totipotential germ cells with concomitant commitment. Teratoma with malignant transformation has been classified into two clinical and pathologic types: (i) induced by chemotherapy or irradiation; and (ii) naturally occurring type.^{2,3} Almost all cases of teratoma with malignant transformation are of the former type, and these tend to occur in young patients with an initial presentation of a malignant germ cell tumour. Naturally occurring teratoma with malignant transformation in the mediastinum is extremely rare, and the risk of malignant transformation may be increased with a long-standing, mature teratoma. Little is known about the incidence and clinicopathological features of somatic-type malignancies in mediastinal mature teratoma because of its rarity.

Only one case of squamous cell carcinoma and three cases of carcinoid, arising sporadically, in mature teratoma of mediastinum have been reported in literature.^{4,5} A review of the previous sporadic case

Table. Observations from published data on mature teratoma with adenocarcinoma (somatic type malignancy) in the mediastinum

Study ^{ref}	Age (in years)	Gender	Symptoms	Site	Tumour Size (cm)	Prognosis after Surgery
Morinaga ²	66	M	None	Anterior	8 x 6.5	30 months, alive
Jung ³	49	M	Haemoptysis	Anterior	12 x 10	6 months, dead
Sakurai ⁴	57	M	None	Anterior	14 x 10	20 months, alive
Shimizu ⁶	43	F	Breathlessness	Anterior	14 x 12	6 months, alive
Popp ⁸	27	M	Breathlessness	Anterior	Large (exact size not mentioned)	Recurrence after few weeks
Chang ⁹	35	M	Back pain	Posterior	20 x 8	2 years, alive
Chen ¹⁰	59	F	Breathlessness	Left hemithorax	18 x 18	Died in accident after 2 chemo cycles
Khurana ¹¹	21	M	None	Anterior	12 x 9.8	6 months, alive
Present case	32	F	Dry cough	Anterior	11.4 x 10.6	2 months, alive

M=Male; F=Female

reports of mature teratoma with adenocarcinoma in the mediastinum is shown in the table.^{2-4,6-9} Although there are no definite clinical characteristics for the diagnosis of a mature teratoma with somatic-type malignancy in mediastinum, these tumours are likely to be seen in older patients with mature teratomas of long duration, in comparison with the common age (early adulthood) for the diagnosis of mature teratoma.¹⁰ The tumour tends to be at least 10cm in size or larger. Adenocarcinoma is likely to be the most frequent histopathological type of somatic-type malignancies, in contrast to ovarian dermoids where the most frequent histopathological type is squamous cell carcinoma (75%), followed by adenocarcinoma and carcinoid.⁶ Areas of thickening in mature teratoma should raise the suspicion of malignancy. However, in our case, no such well-defined thickened areas were seen except for a ridge like elevation and adenocarcinoma was observed in all sections including thin cyst wall as well as ridge like elevation.

Surgical resection plays an essential role in therapy when the tumour is limited to a single site, but the role of chemotherapy has not been adequately addressed. Chemotherapy based on histopathology (such as 5-fluorouracil-based regimen for adenocarcinoma transformation) has recently been advocated by Donadio *et al*¹¹ and it may help improve patient outcome.

In conclusion, the possibility of somatic-type malignancies (non-germ cell malignancies) should be considered while evaluating mature teratomas, especially when a suspicious mature teratoma that is large in size is detected in middle-aged patients, even in the absence of well-defined thickened wall areas.

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