

Case Report

Ectopic hamartomatous thymoma: report of a case and review of literature

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Received July 17, 2015; Accepted August 25, 2015; Epub September 1, 2015; Published September 15, 2015

Abstract: Ectopic hamartomatous thymoma (EHT) is an exceedingly rare lesion that usually arises in the lower neck and mainly affects adult men. We present the clinicopathological features of a case of EHT in a 28-year-old Chinese male, together with a literature review. Ultrasound imaging and a computed tomography (CT) scan of the neck demonstrated a 3.0-cm well-defined nodule of heterogeneous density located within the left sternocleidomastoid muscle. The patient underwent a gross total resection of the tumor. Grossly, the well-demarcated, encapsulated mass had a predominantly solid and gray-white appearance admixed with microcystic foci filled with serous content and yellowish regions. The lesion consisted of an irregular admixture of spindle cells, epithelium, and mature adipose tissue. Immunohistochemistry showed that both the spindle cell and epithelial components were diffuse and had intense nuclear positivity for p63 and cytoplasmic reactivity for pan-cytokeratin, CK7, and CK19. The patient was followed for 18 months without any evidence of metastasis or recurrence.

Keywords: Ectopic hamartomatous thymoma, neck, thymoma, tumor

Introduction

Ectopic hamartomatous thymoma (EHT) is an exceedingly rare neoplasm that usually arises in the lower neck, including the supraclavicular, suprasternal, or presternal areas; on histological examination, EHT characteristically consists of an admixture of spindle cell, epithelial cell, and mature adipose components. Since it was first provisionally described as “a mixed tumor featuring mesenchymal and epithelial components” by Smith and McClure in 1982, 59 cases of EHT have been documented in the English-language literature (**Table 1**) [1-28]. In this study, we describe the clinicopathological and immunohistochemical features of an EHT located in the left supraclavicular region. In addition, we review the available literature on this subject.

Case report

A 28-year-old Chinese male, in otherwise good health, presented with a 1-year history of a

painless mass located in the left supraclavicular region, with slight tenderness on palpation. The tumor had enlarged slowly for approximately 3 months. His past medical history and family history were unremarkable. He did not use alcohol or tobacco. His routine laboratory data were all within normal ranges. Physical examination showed a 3.0-cm, oval, movable lump in the left supraclavicular area. The firm mass was slightly tender and had distinct margins on palpation. Using ultrasound imaging, a 3.0-cm well-defined nodule of heterogeneous density was observed within the left sternocleidomastoid muscle. A computed tomography (CT) scan of neck also revealed an oval heterogeneous mass with mixed components of soft tissue and fat density. The image was not enhanced with contrast administration. There were no abnormalities on the chest X-ray. From the ultrasonography and CT findings, the clinical impression at this time was a schwannoma, lymph node or unusual lipoma. Fine-needle aspiration was performed and yielded the diagnosis of a

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Table 1. Clinicopathological data for reported cases of ectopic hamartomatous thymoma

Reference	Sex/age (Yr)	Location	Size (cm)	Duration before Operation (Yr)	Therapy	Follow-up (Yr)
Smith et al. [1]	F/55	L supraclavicular area	8.0	Many	Local resection	NED, 3
Rosai et al. [2]	M/35	L supraclavicular area	4.0	Long time	Local resection	NED, 8
	M/26	L supraclavicular area	6.0	Long time	Local resection	NED, 5
	M/40	Suprasternal area	2.0	Long time	Local resection	NED, 4
	F/43	R supraclavicular area	5.0	Long time	Local resection	NED, 2
Fetsch et al. [3]	M/79	Superficial chest wall	19.0	30	Local resection	NA
	M/38	Beneath SCM, above medial clavicle	5.5	NA	Local resection	NA
	M/37	Superficial	3.5	1	Local resection	NA
	M/65	Superficial	10.0	30	Local resection	NA
Saeed et al. [4]	M/42	Suprasternal area	5.0	18	Local resection	NA
Chan et al. [5]	M/66	Suprasternal area	7.0	NA	NA	NED, 7
	M/58	Suprasternal area	NA	NA	NA	No recurrence, DUD, 4
	M/47	L supraclavicular area	3.8	NA	NA	NED, 3.5
	M/Adult	Suprdclavicular area	3.0	NA	NA	NA
	M/68	L sternoclavicular area	3.0	NA	NA	Recent case
Armour et al. [6]	M/47	L supraclavicular area	3.8	Many	NA	NA
Doctor et al. [7]	F/49	Suprasternal area	2.4	NA	NA	NA
Michal et al. [8, 9]	M/31	R supraclavicular area	6.0	6/12	NA	NED, 5
	M/39	R supraclavicular area	5.5	NA	NA	NA
	M/38	Suprasternal area	3.0	NA	NA	NED, 0.5
	M/36	Suprasternal area	1.5	NA	NA	NED, 0.5
Eulderink et al. [10]	M/27	Presternal area	3.5	Many	NA	NA
Hirokawa et al. [11]	M/63	Suprasternal area	3.5	NA	Local resection	NA
Michal et al. [12]	M/43	R suprasternal area	3.0	NA	NA	NA
Henderson et al. [13]	M/39	R sternoclavicular area	1.4	NA	NA	NA
Zhao et al. [14]	M/71	L supraclavicular area	9.0	40	Local resection	NA
	M/52	R supraclavicular area	3.5	0.5	Local resection	NED, 3
Marschall et al. [15]	M/22	Supraclavicular area	5.0	7	Local resection	NED, 3
Fukunaga [16]	M/52	L supraclavicular area	1.5	3/12	Local resection	NED, 8/12
Lee et al. [17]	M/59	L supraclavicular area	3.5	4	Local resection	NA
Kazakov et al. [18]	F/71	Interface of the posterior axillary region and back	3.5	30	NA	NA
Fetsch et al. [19]	M/28	L neck	2.0	2-3	NA	2 recurrences at 1 and 13 Yrs postoperatively
	M/29	Suprasternal area	3.5	NA	NA	NA
	M/30	Suprasternal area	7.0	5	NA	NA
	M/35	Chest wall, near sternal notch	7.0	NA	NA	NA

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	M/36	L infraclavicular area	4.0	NA	NA	NA
	M/36	Supraclavicular fossa	5.0	Many	NA	NA
	M/40	L supraclavicular area	6.2	11-12	NA	NED, 13
	M/40	Medial clavicular area	6.5	<1	NA	NA
	M/47	R supraclavicular	4.0	2	NA	NED, 6+2/12
	M/47	L sternoclavicular joint area	2.5	NA	NA	NA
	M/48	L sternoclavicular joint area	2.5	NA	NA	NA
	M/48	L sternoclavicular joint area	2.5	>2	NA	NED, 20+4/12
	M/63	L clavicular region area	2.5	NA	NA	1 recurrence at 2+5/12 postoperatively
	F/64	Suprasternal to manubrium	3.0	<1	NA	Recent case
	M/65	R sternoclavicular joint area	2.5	30	NA	NED, 16+8/12
	M/78	L clavicular area	2.0	2/12	NA	NED, 5
Iida et al. [20]	F/89	L supraclavicular area	6.0	19	Local resection	NED, 11/12
Kushida et al. [21]	M/19	L supraclavicular area	5.0	6/12	Local resection	NA
Sakurai et al. [22]	M/26	L supraclavicular area	2.0	Long time	Local resection	NED, 11
Choi et al. [23]	M/44	L sternoclavicular area	7.5	7	NA	NED, 1+3/12
Weinreb et al. [24]	M/77	L supraclavicular area	4.5	15-20	NA	NA
Shim et al. [25]	M/34	L supraclavicular area	6.0	2/12	Local resection	NA
Liang et al. [26]	F/29	Beneath L SCM	4.0	3	NA	NED, 2+5/12
	M/31	Beneath L SCM	8.0	2	NA	NED, 9/12
	M/47	Anterior chest	4.5	7	NA	NED, 8+4/12
	F/53	R supraclavicular area	8.5	2	NA	NA
Cheng et al. [27]	M/31	R supraclavicular area	8.0	6	Local resection	NED, 3
Huang et al. [28]	M/40	L clavicular area	4.0	0.5/12	Local resection	NED, 2+7/12
Present case	M/28	L supraclavicular area	3.0	1	Local resection	NED, 1.5

Yr: year; F: female; L: left; NED: no evidence of disease; M: male; R: right; NA: not available; SCM: sternocleidomastoid muscle; DUD: died of unrelated disease.

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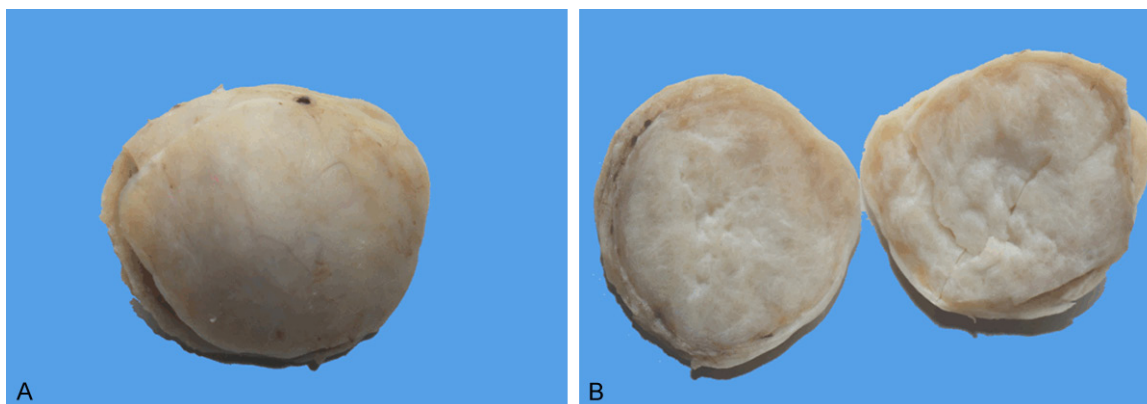
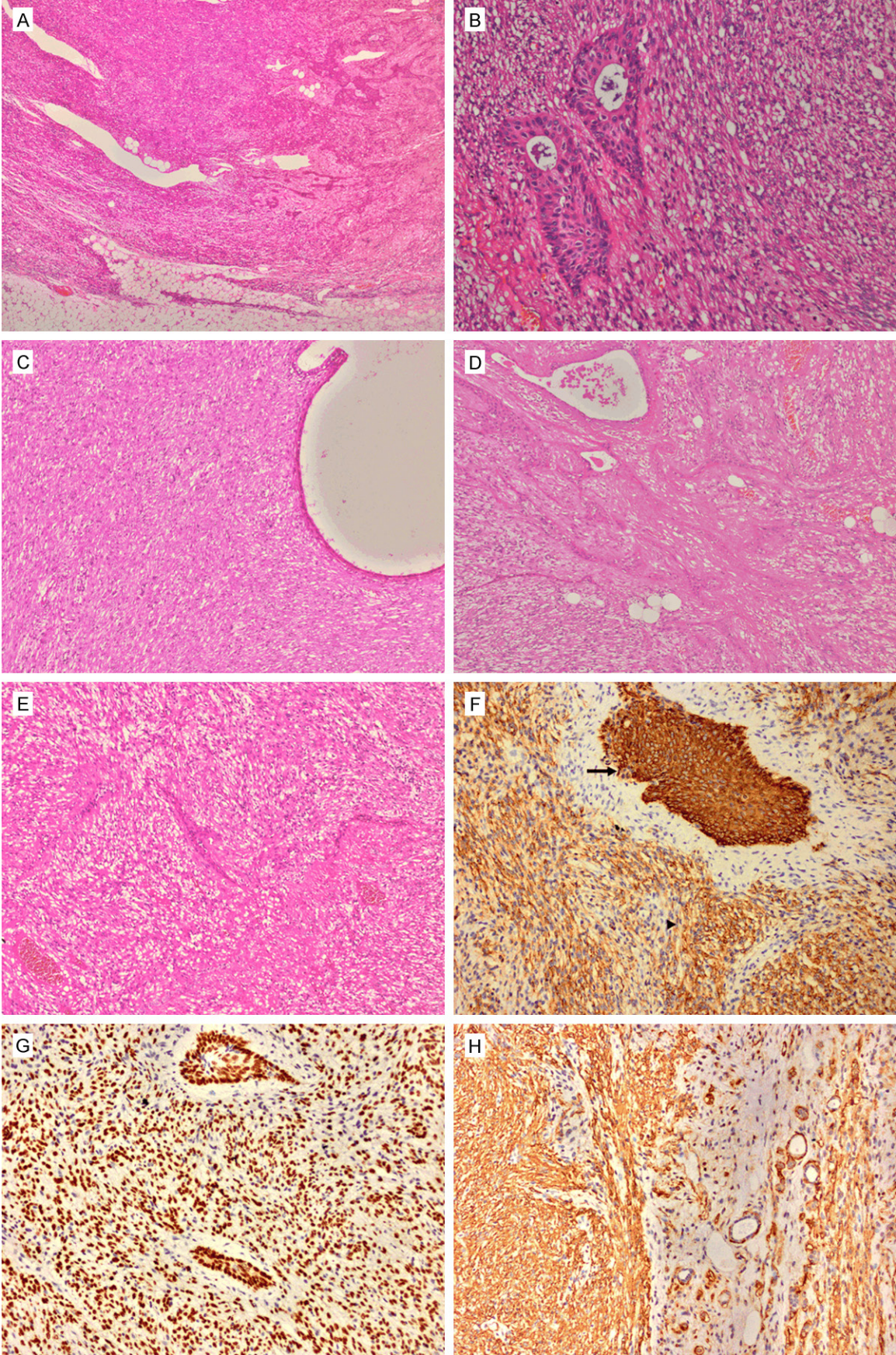


Figure 1. Gross appearance of an ectopic hamartomatous thymoma. A: The tumor was well defined with a smooth external surface. B: The encapsulated tumor has a predominantly solid and gray-white appearance admixed with microcystic foci and yellowish regions on the cut surface.

mesenchymal tumor, categorized as favor benign. The patient subsequently underwent gross total surgical resection of the tumor under field block anesthesia without any grossly residual tumor. During surgery, a well-demarcated oval tumor, sized 3.0 cm × 2.5 cm × 2.0 cm, was located in the middle-inferior portion of the left sternocleidomastoid muscle without invasion. The tumor resulted in intermediately dorsal shifts of both the left jugular vein and carotid artery owing to its pressure. However, no evidence of tumor invasion into the adjacent tissues was present. The tumor was not attached to the thyroid, clavicle, or mediastinum. The patient's postoperative course was uneventful. He was well, without evidence of recurrence or metastasis, 18 months after resection. On gross pathology, the well-defined oval tumor was 3.0 cm × 2.5 cm × 2.0 cm in size with a smooth external surface (**Figure 1A**). Sectioning revealed an encapsulated mass with a predominantly solid and gray-white appearance admixed with microcystic foci filled with serous content and yellowish regions, most of which were located in the periphery of the tumor (**Figure 1B**). The tumor was moderately firm and rubbery in consistency. Both hemorrhage and necrosis were absent. Microscopic examination revealed that the obviously encapsulated tumor was relatively cellular and consisted of an irregular admixture of three distinct components: spindle cells, epithelium, and mature adipose tissue, in variable proportions (**Figure 2A**). On low-power microscopic examination, the spindle cell elements dominated the microscopic field and accounted for

70% of the entire lesion, followed by the epithelial component and adipose tissue, which comprised 20% and 10% of the tumor, respectively. The spindle cells were mainly arranged in haphazard and vague fascicular patterns, but occasionally, a storiform structure was present. Although most areas of spindle cells were cellular, a small number of regions with sparse spindle cells or hyaline degeneration were noted; these were generally adjacent to some epithelial nests, mature adipose tissue, or dilated vessels. The bland-appearing, medium-sized spindle cells possessed moderately eosinophilic, ill-defined cytoplasm and vesicular nuclei with small or inconspicuous nucleoli. The nuclei were oval to spindle in shape and had fine chromatin and small or inconspicuous nucleoli (**Figure 2B**). The polygonal epithelium mainly consisted of primitive squamous, squamous, cuboidal, flattened, and columnar cells that were arranged in small, solid nests with irregularly branching cords, and cysts with variable size. The nonkeratinizing squamous epithelium formed nested structures, some of which had cystic degeneration (**Figure 2B**). The branching corded structures comprised primitive squamous epithelial cells and were sometimes connected with squamous cell nests or cysts. The luminal layer of the tubular structures was lined by cuboidal or flattened epithelium. The cysts had a single layer of luminal lining cells with variable appearances, ranging from flattened and cuboidal to columnar cells (**Figure 2C**). Intraluminal inspissated eosinophilic material was identified in some cystic structures (**Figure 2D**). There was a myoepithe-

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Figure 2. Microscopic and immunohistochemistry findings of the tumor. A: The neoplasm comprised an admixture of spindled cells, epithelial cells, and mature adipose tissue (hematoxylin and eosin stain, original magnification, $\times 40$). B: Nonkeratinizing squamous epithelium with cystic degeneration and bland spindle cells in which no pleomorphism, abnormal mitosis, or nuclear atypia were present (hematoxylin and eosin stain, original magnification, $\times 200$). C: A cystic structure lined by flattened, cuboidal, or columnar epithelial cells (hematoxylin and eosin stain, original magnification $\times 100$). D: Branching corded structures of epithelium and cystic structures with inspissated eosinophilic material (hematoxylin and eosin stain, original magnification, $\times 100$). E: Gradual transition from branching cords of the epithelium to spindle cells (hematoxylin and eosin stain, original magnification, $\times 100$). F: Both the spindle cell and epithelial components were positive for pan-cytokeratin, and the level of immunostaining of the latter (arrow) was stronger than that in the former (arrowhead) (original magnification, $\times 200$). G: Strong and diffuse P63 immunoreactivity was present in epithelial cells and spindle cells (original magnification, $\times 200$). H: Immunohistochemical staining for SMA was positive in the spindle cells and myoepithelial cells of the tubular and cystic structures but negative in the epithelial cells (original magnification, $\times 200$).

lial layer around the circumference of the tubular and cystic structures. Although the epithelium, especially in the cystic and squamous cell nested areas, was well separated from the spindle cell component, a gradual transition from branching cords and strands of epithelial cells to spindle cells was frequently observed (**Figure 2E**). Mature fat cells and lymphocytes were scattered throughout the lesion. The former were in various proportions in different areas of the tumor, such as in clusters and foci of adipocytes, but had a tendency to be predominantly distributed at the periphery of the tumor. The diffuse but sparse infiltration of small lymphocytes was present mostly in the background of the spindle cell component. Atypical or malignant features such as nuclear atypia, cellular pleomorphism, vascular and capsular invasion, and necrosis were absent from the entire lesion. Mitoses were scarce, with 1.4 per 50 high-power fields (HPFs), but no abnormal mitoses were observed. Immunohistochemical staining demonstrated that both the spindle cell and epithelial components were diffuse and intense cytoplasmic reactivity for pan-cytokeratin (**Figure 2F**), CK7, and CK19, and nuclear positivity for p63 (**Figure 2G**). Moreover, the extent of reactivity for pan-cytokeratin, CK7, and CK19 in epithelial components was stronger than in the spindle cell portion. The spindle cells were also strongly and uniformly positive for smooth muscle actin (SMA) (**Figure 2H**) but focally and weakly reactive for vimentin and CD99. The epithelial cells were negative for CD99, vimentin, and SMA; however, the outer layer cells of the cystic and tubular structures were positive for SMA and vimentin, which corresponded to the myoepithelial cell position (**Figure 2H**). Staining for S-100 protein, epithelial membrane antigen (EMA), desmin, Myo-D1, and HMB45 was negative in the spindle cell and epithelial compo-

nents. The scattered intratumoral lymphoid component was positive for CD45RO and CD3 but negative for CD20, CD79a, CD1a, and CD99. MIB-1 stained only approximately 1.5% of the spindle and epithelial cells, with 1000 cells counted. These overall clinicopathological features found in the patient met the diagnostic criteria for EHT.

Discussion

An EHT, which was first formally described as a new entity in the 2002 edition of the WHO classification of soft-tissue tumors, is an extremely rare benign tumor characteristically comprising an admixture of spindle cells, epithelial islands, and mature adipose tissue [29]. It was first described by Smith and McClure in 1982 and subsequently was designated as an “ectopic hamartomatous thymoma” by Rosai et al. in 1984 [1, 2].

So far, a total of 60 pathologically confirmed cases of EHT have been reported in the English literature, including our patient (**Table 1**) [1-28]. As documented in **Table 1**, EHT is usually a slowly growing mass with a long duration, ranging from 15 days to 40 years, prior to surgery. EHT often occurs in middle-aged adults (age range: 19 to 89 years; mean: 46.1 years). The male-to-female ratio is 52:8, demonstrating the remarkable male predominance. The most common locations of EHT are the supraclavicular, suprasternal, and sternoclavicular areas. Other sites more rarely affected include the chest, the interface of the posterior axillary region and back, and the presternal, infraclavicular, and clavicular areas. In the 58 cases of EHT that documented tumor size, the tumors ranged from 1.4 cm to 19.0 cm (mean, 4.8 cm), and the follow-up interval of the 30 cases of EHT with available follow-up information ranged

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from 0.5 years to 13 years (mean: 5.2 years). All patients with available follow-up information were alive except one who had died 4 years postoperatively of unrelated causes. One patient's tumor recurred once at a post-operative interval of 2 years and 5 months, and one patient relapsed twice at 1 year and 13 years after surgery. However, the recurrences of two patients reported by Fetsch et al. were due to incomplete local excision, and both patients had no evidence of disease for a long time after a complete re-excision [19].

It is well known that nearly all of EHTs show no evidence of atypical or malignant features and pursue a benign clinical course, and complete local excision is the first treatment of choice. The malignant epithelial portions, consisting of a dysplastic glandular component that is usually arranged in a cribriform structure bridging several glandular configurations, were observed in 2 cases of EHT reported by Michal et al. [8, 9]. As a result, some authors have advocated that the probability of the malignant convert of tumor cells within EHT must be considered and that sufficient sampling and careful assessment of the malignant component in each instance of EHT are essential [19, 22]. However, no adverse outcome was documented in one of the two patients with an EHT with a malignant epithelial component; there was no documented follow up on the other patient with an EHT with a malignant epithelium [8, 9].

Myoid cells were observed among the spindle cell areas in variable proportions in three cases of EHT, which immunohistochemically were myoglobin positive and desmin negative [4, 6, 14]. Interestingly, immunostaining for cytokeratin and EMA was positive in the myoid component in one case of EHT [14] and negative in the remaining two cases [4, 6]. Occasional poorly formed cross-striations were demonstrated in such cells in one case by phosphotungstic acid hematoxylin (PTAH) staining [4].

The exact origin of EHT remains controversial. The prevailing original view is that this tumor originates in the thymic anlage; possible sources include the third and fourth pharyngeal pouches, the cervical sinus of His, and the ultimobranchial body [1-4, 6]. However, many characteristics of this lesion have challenged the hypothesis of thymic origin. Residual thymic structures have never been described in asso-

ciation with EHT and a mediastinal counterpart was absent in all cases of EHT reported to date [8, 9, 13, 15, 19]. Furthermore, the reported unusual locations of EHT, including the pre-sternal and dorsal locations, could also exclude origin in a remnant of thymic tissue because of the scarcity of thymic remnants in both the pre-sternal and dorsal areas [10, 18]. Considering the adenocarcinomatous differentiation, myo-epithelial cell component, and clusters of gland acini component in EHT, Michal et al. believed that there could be salivary gland differentiation in EHT [8]. In 2000, Henderson and Gupta proposed an EHT origin from the branchial pouch endoderm rather than from thymic remnants [13]. After analyzing the clinicopathological and immunohistochemical features of 21 cases of so-called "ectopic hamartomatous thymoma", Fetsch et al. suggested that the nomenclature "branchial anlage mixed tumor" more accurately reflects the nature of this rare lesion [19]. Weinreb et al. reported a case of EHT with skin adnexal differentiation with immunoreactivity for EMA, androgen, and BRST-2 and suggested that "a mixed tumor of skin adnexal origin" is another possible designation for EHT [24].

The pathologic differential diagnosis of EHT includes biphasic synovial sarcoma, malignant schwannoma with epithelial differentiation, a mixed tumor of skin or salivary gland tissue, and thymolipoma. Briefly, the presence of a well-defined lesion usually located in the lower neck with characteristic spindle cell and epithelial cell components and mature adipose tissue, together with the immunohistochemical features of both the spindle cell and epithelial cell components having cytokeratin positivity should facilitate an accurate diagnosis of EHT. Biphasic synovial sarcoma can easily be excluded because it rarely shows squamous differentiation and the spindle tumor cells have higher cellularity, greater nuclear hyperchromasia, and mitotic figures. Furthermore, the spindle cell component in synovial sarcoma shows foci of patchy positivity for cytokeratin [30]. Tumor cells in malignant schwannoma with epithelial differentiation are S-100 protein positive and cytokeratin negative. In contrast, tumor cells in EHT are cytokeratin positive and S-100 protein negative [31]. Differentiating a mixed tumor of skin or salivary gland tissue from an EHT is based on the former's chondromyxoid stroma

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and immunoreactivity for S-100 protein, which are absent in the latter [32, 33]. Thymolipoma could be excluded, as it nearly always arises in the mediastinum rather than in the neck, and lacks the prominent spindle cell component of EHT [34].

Conclusion

In conclusion, we have reported a case of EHT in a 28-year-old male and presented a review of the literature to clarify the clinical and pathological characteristics of this rare tumor. EHT is a rare lesion with characteristic histological and immunohistochemical features and is frequently located in the lower neck area. Correct identification of this unique and distinctive neoplasm is important because the tumor shows a benign biologic behavior and the therapy, if not complicated by a malignant component, is local excision. More cases of EHT must be studied to draw definitive conclusions about the tumor's exact histogenesis.

Acknowledgements

The authors thank American Journal Experts for editing the present paper. This work was supported by the Natural Science Foundation of Shandong Province (Grant No. ZR2012HM-018).

Disclosure of conflict of interest

None.

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