

## Short Communication



# A Rare Case of Myxoid Liposarcoma Combined With Papillary Carcinoma in the Same Thyroid Gland

Jung Hyun Moon <sup>1,\*</sup>, Jun Sung Lee <sup>2,\*</sup>, Su-Jin Shin <sup>3</sup>, Jin Seok Lee <sup>2</sup>, Hyeok Jun Yun <sup>2</sup>, Yong Sang Lee <sup>2</sup>, Hang-Seok Chang <sup>2</sup>

<sup>1</sup>Yonsei University College of Medicine, Seoul, Korea

<sup>2</sup>Department of Surgery, Thyroid Cancer Center, Gangnam Severance Hospital, Institute of Refractory Thyroid Cancer, Yonsei University College of Medicine, Seoul, Korea

<sup>3</sup>Department of Pathology, Gangnam Severance Hospital, Yonsei University College of Medicine, Seoul, Korea

## OPEN ACCESS

Received: Nov 29, 2022

Revised: Dec 2, 2022

Accepted: Dec 7, 2022

Published online: Dec 14, 2022

### Correspondence to

Yong Sang Lee

Department of Surgery, Thyroid Cancer Center, Gangnam Severance Hospital, Institute of Refractory Thyroid Cancer, Yonsei University College of Medicine, 211 Eonju-ro, Gangnam-gu, Seoul 06273, Korea.  
Email: medilys@yuhs.ac

\*Jung Hyun Moon and Jun Sung Lee contributed equally to this article as a first author.

Copyright © 2022. Korean Association of Endocrine Surgeons; KAES  
This is an Open Access article distributed under the terms of the Creative Commons Attribution Non-Commercial License (<https://creativecommons.org/licenses/by-nc/4.0/>).

### ORCID iDs

Jung Hyun Moon   
<https://orcid.org/0000-0001-7178-5069>  
Jun Sung Lee   
<https://orcid.org/0000-0002-4727-3028>  
Su-Jin Shin   
<https://orcid.org/0000-0001-9114-8438>  
Jin Seok Lee   
<https://orcid.org/0000-0001-9755-9801>  
Hyeok Jun Yun   
<https://orcid.org/0000-0001-6004-0782>  
Yong Sang Lee   
<https://orcid.org/0000-0002-8234-8718>  
Hang-Seok Chang   
<https://orcid.org/0000-0002-5162-103X>

## ABSTRACT

Myxoid liposarcoma is a rare soft-tissue sarcoma that is usually observed in the retroperitoneum and extremities. In this case report, we describe a 39-year-old female patient who presented with myxoid liposarcoma of the thyroid and papillary thyroid carcinoma with recurrent lesions in the lung and retroperitoneum.

**Keywords:** Thyroid; Myxoid liposarcoma; Papillary carcinoma; recurrence

## INTRODUCTION

Sarcomas are a group of heterogeneous tumors with genetic abnormalities. Genetic abnormalities range from common chromosomal amplification or loss to more complex rearrangements such as multiple chromosomes. Liposarcoma is one of the most common soft tissue sarcomas in adults. It arises from the precursor of adipocytes and is mainly found in the retroperitoneum and extremities, but it is also found in lesions of the thighs, torso, head, and neck (1). Liposarcoma has three main morphological subgroups, one of which is myxoid/round liposarcomas (2).

About 23%–58% of patients with soft-tissue sarcoma exhibit metastatic lesions after undergoing treatment for primary tumors (3). Most of them are lung metastases, but in cases of myxoid liposarcomas, extrapulmonary metastases, such as those in the retroperitoneum, abdomen, and bone, are also common (4).

In this report, we describe a rare case of myxoid liposarcoma of the thyroid with concurrent papillary thyroid carcinoma.

## CASE DESCRIPTION

A 39-year-old female patient with hypertension and diabetes presented with a large and rapidly growing thyroid mass. The patient noted a thyroid mass 7 years prior but had not been

**Conflict of Interest**

No potential conflict of interest relevant to this article was reported.

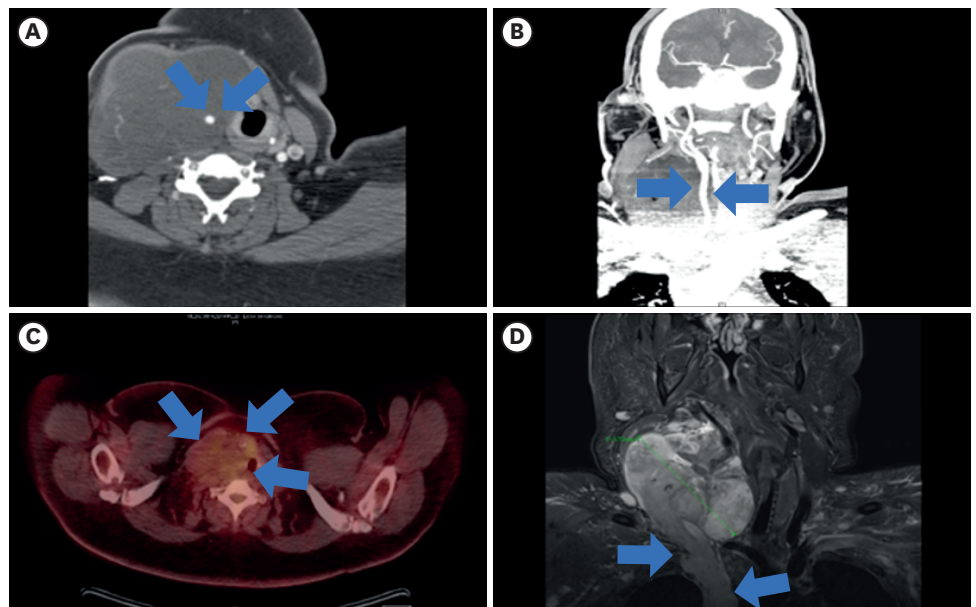
**Author Contributions**

Conceptualization: Yong Sang Lee; Writing - original draft: Jung Hyun Moon, Jun Sung Lee; Writing - review & editing: Jun Sung Lee, Yong Sang Lee.

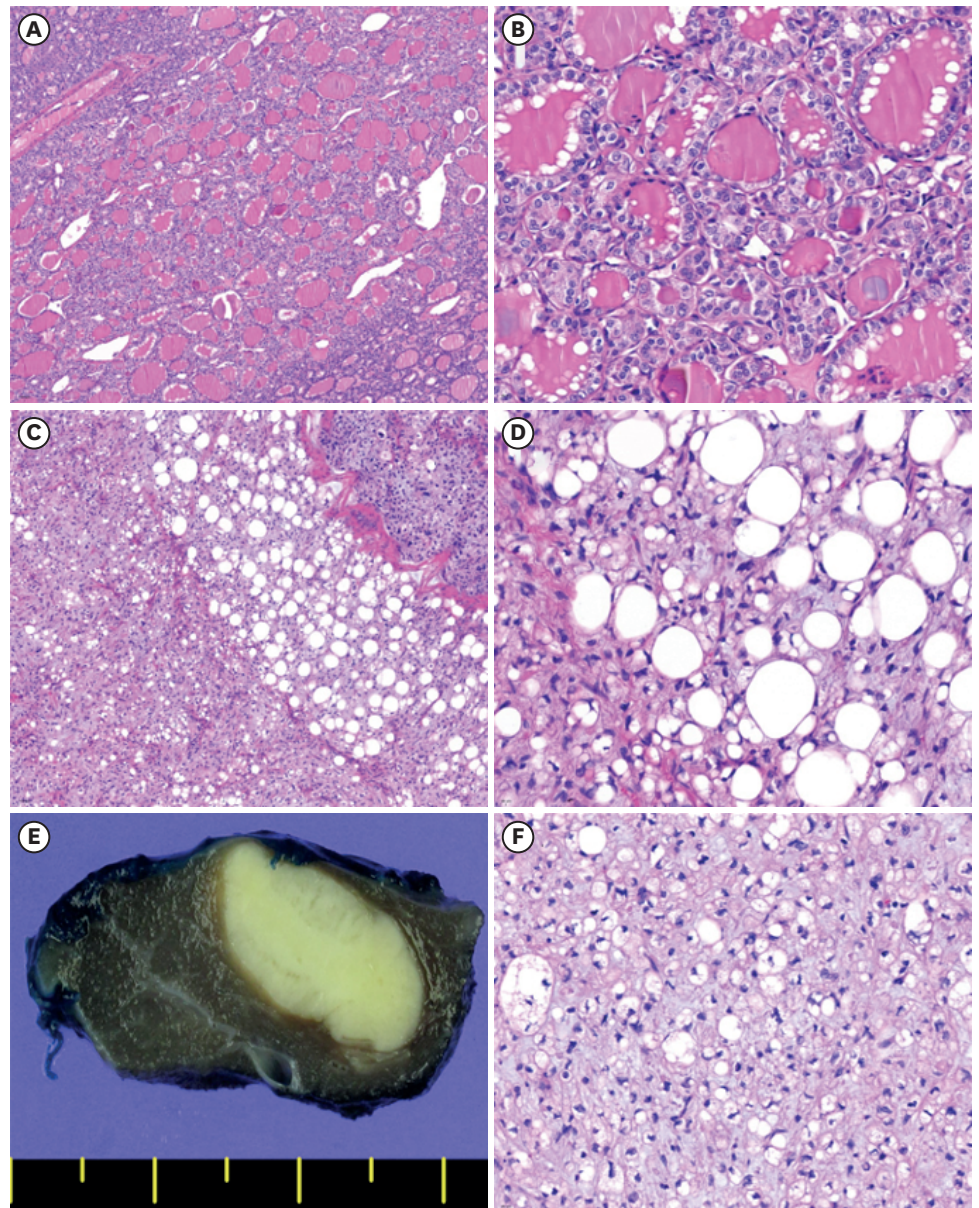
treated. The mass grew rapidly 3 months before visiting the hospital. The patient exhibited no symptoms of a growing thyroid mass.

Thyroid function tests were normal, except for a thyroglobulin Ag of 77.39 ng/mL (0.04 to 32.5 ng/mL). Ultrasonography, computed tomography (CT), positron emission tomography (PET), and magnetic resonance imaging (MRI) revealed a well-defined heterogeneous mass of approximately 11.5×11.0 cm in size in the right anterior cervical space involving the right thyroid gland and extending along the right internal jugular vein and subclavian vein. Several enlarged lymph nodes were observed around the primary mass lesion (**Fig. 1**). CT revealed a large thyroid mass of approximately 11×10 cm in size that encased the right common carotid artery (A & B). PET revealed a lobulated low-density mass of approximately 12×7 cm in size with mild FDG uptake in the right eye. The lobe of thyroid (C) and MRI revealed a well-defined T2 high, heterogenous enhancing mass lesion of 11.5×11.0 cm size in the right anterior cervical space involving the right thyroid gland. The tumor extended to the right internal jugular vein to the superior vena cava (D).

Bilateral total thyroidectomy with central compartment node dissection and right modified radical neck dissection was safely performed. The main mass around the right thyroid gland was dissected, and the right recurrent laryngeal nerve and right internal jugular vein were sacrificed because of the huge tumor encasing them. Fortunately, the right common carotid artery and right vagus nerve were preserved. Mass was left a little because it was impossible to completely resect around right common carotid artery (R1 resection). Frozen sections of a large mass demonstrated atypical short spindle cells with myxoid stroma, suggestive of a malignant mesenchymal tumor. The final pathological diagnosis was papillary carcinoma of the right thyroid gland and soft tissue attached to the right lobe as a myxoid liposarcoma (**Fig. 2**). The papillary carcinoma was follicular and solid pattern, intra-thyroidal lesion with



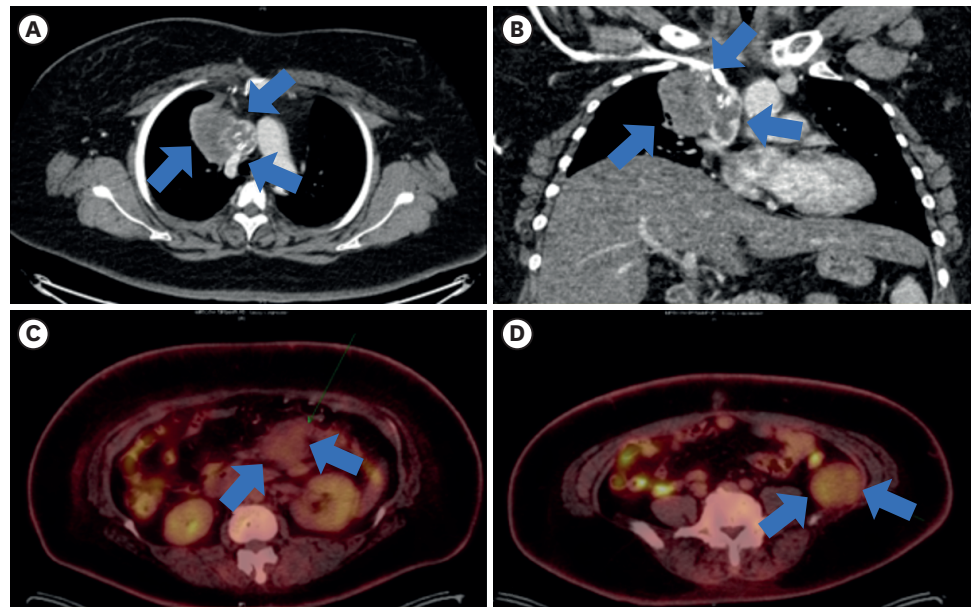
**Fig. 1.** Imaging study of thyroid tumor before surgery. Computed tomography revealed a large thyroid mass of approximately 11×10 cm in size that encased the right common carotid artery (A, B). Positron emission tomography revealed a lobulated low-density mass of approximately 12×7 cm in size with mild FDG uptake in the right lobe of the thyroid (C), and magnetic resonance imaging revealed a well-defined T2 high, heterogenous enhancing mass lesion of 11.5×11.0 cm in size at the right anterior cervical space involving the right thyroid gland. The tumor extended along the right internal jugular vein to the superior vena cava (D).



**Fig. 2.** Histologic features of papillary thyroid carcinoma, follicular pattern (A, B), myxoid liposarcoma (C, D) and gross picture and histologic figures of metastatic myxoid liposarcoma (E, F).

3.0 cm maximum diameter of right thyroid upper to mid pole. The myxoid liposarcoma was mass that fragmented into two part (8.5×8.0×6.0 cm and 5.5×2.0×2.0 cm), attached to right thyroid. Though we sacrificed the right recurrent laryngeal nerve, the voice of patient didn't change much compared to before surgery. Perhaps this is because the nerve function was already almost gone before surgery. After the surgery, we performed the treatment for each of papillary carcinoma and myxoid liposarcoma separately.

The patient underwent adjuvant radiotherapy with a total of 56 Gy in 28 fractions to control the remaining liposarcoma after the surgery. There were no complications during the treatment, and the patient tolerated the treatment well.

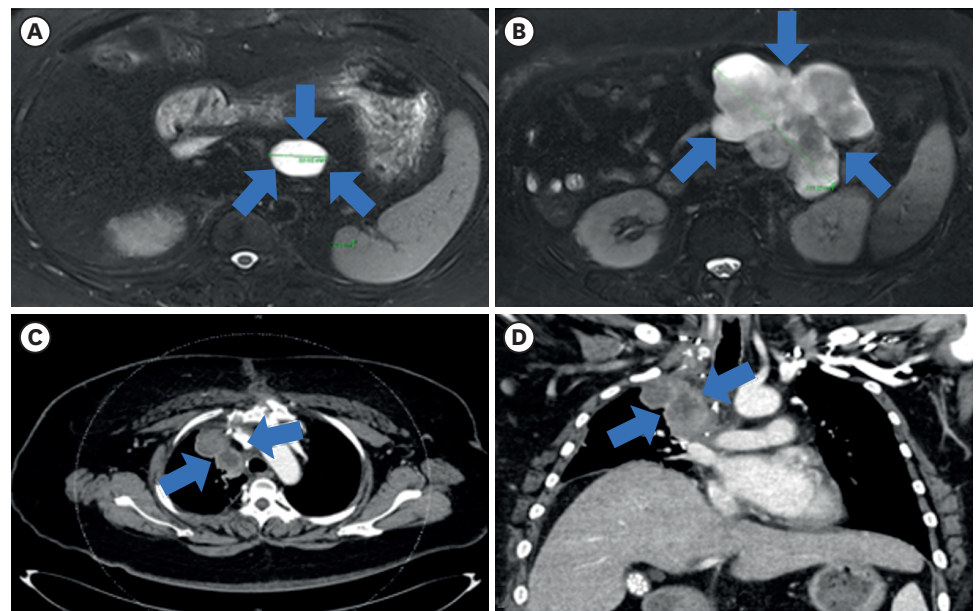


**Fig. 3.** Imaging of the right lung revealing a metastatic lesion and abdominal metastasis. Chest computed tomography revealed an enhancing metastatic lesion in the right lung, right pleura, and right upper paratracheal area with an invasion of the superior vena cava (A, B), and positron emission tomography revealed additional metastatic lesions in the left side of the mesentery and left retroperitoneum (C, D).

A year after the patient underwent adjuvant radiotherapy, chest CT revealed a newly developed tiny nodule in the right middle and lower lobes. In the follow-up evaluation 6 months later, chest CT and PET revealed a large enhancing mass in the right thorax area with an invasion of the superior vena cava and metastatic lesions in the right lung and mediastinum (**Fig. 3**), and the patient presented with dyspnea and edema in the right arm and on the face. In addition, chest CT revealed an increased metastatic lesion in the right lung, right pleura, and right upper paratracheal area with an invasion of the superior vena cava (A & B), and PET revealed additional metastatic lesions at the left mesentery and left retroperitoneum (C & D).

The patient was hospitalized to undergo surgery for lung lesions. Excision of the pleural mass with the reconstruction of the SVC and wedge resection of the lung was performed for the lung mass. The pathology of the pleural mass demonstrated that it was myxoid liposarcoma, and several lung lesions demonstrated the same results as metastatic liposarcoma.

After the surgery, the tumors observed in the abdomen appeared to grow in size on follow-up imaging (**Fig. 4**). MRI revealed an 11-cm lobulated mass in the mesentery of the upper abdomen consisting of fluid with enhancing solid and fatty components, suggesting liposarcoma with a 2.3-cm mass in the second portion of the duodenum (A & B). Additionally, recurrence was observed in the area where excision of the pleural mass was performed on chest CT (C & D). Therefore, it was thought that surgical treatment would not be helpful to the patient, and the patient's condition was not suitable to endure major surgery, so it was decided to proceed with chemotherapy without surgery. Accordingly, palliative chemotherapy was initiated on February 16, 2022, and the patient is currently being followed up in outpatient clinics.



**Fig. 4.** Imaging of the abdomen after lung surgery revealing abdominal metastasis. Magnetic resonance imaging revealed an 11-cm lobulated mass in the mesentery of the upper abdomen consisting of fluid with enhancing solid and fatty components, suggesting liposarcoma with a 2.3-cm mass in the second portion of the duodenum (A, B). Recurrence in the area where excision of the pleural mass was performed was observed in the chest computed tomography images (C, D).

## DISCUSSION

Liposarcoma is one of the most common forms of soft-tissue sarcoma in adults (2). Sarcomas are rare, accounting for less than 1% of adult malignancies (5). Liposarcomas can be divided into several subtypes, including well-differentiated, myxoid/round-cell, and pleomorphic liposarcomas.

Myxoid liposarcoma usually occurs in the extremities, whereas most liposarcomas prefer the retroperitoneum. Myxoid liposarcoma occurs mainly in those at a young age, from about 35–55 years (6). The prognosis of myxoid liposarcoma depends on the percentage of round-cell liposarcomas, but the exact threshold levels remain controversial (2). The lungs and bones are the most common sites of metastasis for liposarcoma, but myxoid liposarcoma exhibits unusual characteristics of spreading to extrapulmonary soft tissue sites.

Myoid liposarcoma is more sensitive to radiation therapy than the other histological subtypes (7). This can be helpful in cases of tumors invading critical structures, such as vessels, nerves, and bones. However, the local recurrence risk of myxoid liposarcoma is relatively low, ranging from 10%–15% in 3 years, depending on the round-cell component (8). Therefore, adjuvant radiation therapy may be unnecessary in normal cases due to the low risk of local recurrence; however, in this case, the patient underwent adjuvant radiation therapy because of the huge mass invading the major neck vessels. There were no signs of local tumor recurrence after the patient underwent radiotherapy.

However, the need for postoperative adjuvant chemotherapy remains controversial. One study demonstrated that it is difficult to prove that chemotherapy has a clinical benefit (9). However, when it comes to high-risk lesions, such as round-cell liposarcomas or tumors

with a large size, ifosfamide- or doxorubicin-based chemotherapy demonstrates improved clinical outcomes (10,11). In this case, the pathological diagnosis of myxoid liposarcoma of the thyroid did not exhibit any round-cell components, but unfortunately, metastatic lesions were observed.

Compared with its low local recurrence rate, the metastatic potential of myxoid liposarcoma is relatively high, ranging from 23%–58% depending on the round-cell composition (3). Among the several treatment options used to treat myxoid liposarcoma, surgical resection is considered in cases of oligometastasis. In chemotherapy, doxorubicin and ifosfamide exhibit more than a partial response in 40% of patients. Radiotherapy remains an effective treatment option, exhibiting a 100% pathologic treatment response (7). In this case report, the patient experienced metastasis to the lung, mediastinum, mesentery, and retroperitoneum. Surgical resection was performed for metastasis to the lung and mediastinum, and palliative chemotherapy was used to treat the remaining lesions.

Noticeably, thyroid myxoid liposarcoma, which is a very rare disease, was observed along with papillary thyroid carcinoma. There are several possible hypotheses for explaining this finding. First, both myxoid liposarcoma and papillary carcinoma are primary cancer lesions. Two tumors may have accidentally developed in the same lesion or nearby lesions. The coexistence of papillary carcinoma and soft-tissue sarcomas has been previously reported (12).

Another hypothesis is that myxoid liposarcoma, which is considered primary cancer, is actually a metastatic lesion from a different site. A previous case report described an 86-year-old white female patient with a collision tumor of the thyroid with metastatic liposarcoma from the thigh and papillary thyroid carcinoma (13). The abdominal lesions observed in the patient, which were thought to be recurrence, may have been primary cancer, considering that liposarcoma prefers retroperitoneal lesions more than neck lesions, as previously mentioned (1).

In conclusion, sarcoma is very rare that less than 1% of adult malignancies, and we experienced a rare case of myxoid liposarcoma combined with papillary carcinoma in thyroid gland. Clinicians should keep in mind the possibility of other rare types of cancer rather than the papillary thyroid carcinoma.

## REFERENCES

1. Conyers R, Young S, Thomas DM. Liposarcoma: molecular genetics and therapeutics. *Sarcoma* 2011;2011:483154.  
[PUBMED](#) | [CROSSREF](#)
2. WHO Classification of Tumours Editorial Board. World Health Organization Classification of Soft Tissue and Bone Tumours. 5th ed. Lyon: IARC Press; 2020.
3. Kilpatrick SE, Doyon J, Choong PF, Sim FH, Nascimento AG. The clinicopathologic spectrum of myxoid and round cell liposarcoma. A study of 95 cases. *Cancer* 1996;77:1450-8.  
[PUBMED](#) | [CROSSREF](#)
4. Moreau LC, Turcotte R, Ferguson P, Wunder J, Clarkson P, Masri B, et al. Myxoidcell liposarcoma (MRCLS) revisited: an analysis of 418 primarily managed cases. *Ann Surg Oncol* 2012;19:1081-8.  
[PUBMED](#) | [CROSSREF](#)
5. Miller KD, Fidler-Benaoudia M, Keegan TH, Hipp HS, Jemal A, Siegel RL. Cancer statistics for adolescents and young adults, 2020. *CA Cancer J Clin* 2020;70:443-59.  
[PUBMED](#) | [CROSSREF](#)

6. Alaggio R, Coffin CM, Weiss SW, Bridge JA, Issakov J, Oliveira AM, et al. Liposarcomas in young patients: a study of 82 cases occurring in patients younger than 22 years of age. *Am J Surg Pathol* 2009;33:645-58.  
[PUBMED](#) | [CROSSREF](#)
7. Pitson G, Robinson P, Wilke D, Kandel RA, White L, Griffin AM, et al. Radiation response: an additional unique signature of myxoid liposarcoma. *Int J Radiat Oncol Biol Phys* 2004;60:522-6.  
[PUBMED](#) | [CROSSREF](#)
8. Cahlon O, Brennan MF, Jia X, Qin LX, Singer S, Alektiar KM. A postoperative nomogram for local recurrence risk in extremity soft tissue sarcomas after limb-sparing surgery without adjuvant radiation. *Ann Surg* 2012;255:343-7.  
[PUBMED](#) | [CROSSREF](#)
9. Chowdhry V, Goldberg S, DeLaney TF, Cote GM, Chebib I, Kim J, et al. Myxoid liposarcoma: treatment outcomes from chemotherapy and radiation therapy. *Sarcoma* 2018;2018:8029157.  
[PUBMED](#) | [CROSSREF](#)
10. Eilber FC, Eilber FR, Eckardt J, Rosen G, Riedel E, Maki RG, et al. The impact of chemotherapy on the survival of patients with high-grade primary extremity liposarcoma. *Ann Surg* 2004;240:686-95.  
[PUBMED](#) | [CROSSREF](#)
11. Patel SR, Burgess MA, Plager C, Papadopoulos NE, Linke KA, Benjamin RS. Myxoid liposarcoma. Experience with chemotherapy. *Cancer* 1994;74:1265-9.  
[PUBMED](#) | [CROSSREF](#)
12. Ubayama Y, Gotoh M, Yamawaki S, Isu K, Imamura A, Miyakawa A, et al. Four case reports of sarcoma combined with carcinoma. *Gan No Rinsho* 1984;30:827-33.  
[PUBMED](#)
13. Brandwein-Gensler M, Urken M, Wang B. Collision tumor of the thyroid: a case report of metastatic liposarcoma plus papillary thyroid carcinoma. *Head Neck* 2004;26:637-41.  
[PUBMED](#) | [CROSSREF](#)