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Case Report

A Rare and Unusual Association of a Pancoast-Tobias Tumor and Aggressive Vertebral Angiomas

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Abstract: ***Introduction:*** Pancoast-Tobias tumor (PTT), is a rare presentation of lung cancer that affects less than 5% of patients. Vertebral hemangioma is a benign tumor that develops in the endothelial lining of blood vessels. It is rarely symptomatic in adults, with only 0.9 to 1.2% of all vertebral hemangiomas being symptomatic. ***Materials and Methods:*** A 69 years old patient presented with right-sided chest pain, cough but no dyspnea associated with a weight loss of 10 kg over the past 06months. The patient reported inflammatory back pain that had been evolving for 2 years and had worsened in recent months. Chest X-ray-CT scan and MRI revealed a tumor mass in the apical segment of the right upper lobe extending into the right posterior mediastinal space at the costo-vertebral groove, measuring 38*33 mm with D2 and D3 vertebral body nodule in T1 isosignal, T2 hypersignal enhancing after gadolinium injection, consistent with an aggressive vertebral angioma (AVA). A multidisciplinary approach was organized, and the indication was to perform preoperative embolization, followed by hemivertebrectomy of D12 associated with costolobectomy. The patient refused to continue the treatment and passed away after 05 months. ***Discussion:*** Most vertebral angiomas are discovered incidentally and should not be treated. Pain is generally the most common symptom of discovery. MRI remains the most effective in exploring vertebral angiomas. PTT can extend to the cervicothoracic outlet, the diagnosis of these tumors calls for chest X-ray. Thoraco-abdomino-pelvic CT scan is part of the extension assessment and MRI allows for precise analysis. The management of the exclusive association of PTT and AVA remains a a challenge. ***Conclusion:*** The combination of PTT and a set of AVA is extremely rare and doesn't appear to be described in the literature. The central problem remains the management, which is a challenge for the responsible surgical team.

Keywords: Pancoast-Tobias, Tumor, Aggressive, Vertebral, Angiomas, Spine.

INTRODUCTION

Pancoast-Tobias tumor (PTT), also known as the pulmonary apex tumor, is a rare presentation of lung cancer that affects less than 5% of patients [1].

Vertebral hemangioma is a benign tumor that develops in the endothelial lining of blood vessels. It is rarely symptomatic in adults, with only 0.9 to 1.2% of all vertebral hemangiomas being symptomatic [2].

When hemangiomas occur in the thoracic vertebrae, they are more likely to be symptomatic due to the narrowness of the vertebral canal, requiring more aggressive management and close monitoring to prevent neurological sequelae [3, 4].

The combination of PTT and a set of aggressive vertebral angiomas (AVA) is extremely rare and does not appear to be described in the literature. The central problem remains the management, which is a challenge for the responsible surgical team.

CASE PRESENTATION

Our case is about a 69 years old patient

- Medical History: Hypertension and poorly controlled diabetes
- Smoking History: 30 pack-years
- Clinical Presentation:

The patient presented with right-sided chest pain that had been progressing for 4 months, recently

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radiating to the right shoulder, accompanied by a cough but no dyspnea. The overall clinical picture was marked by general deterioration, with a weight loss of 10 kg over the past 6 months.

In addition to thoracic symptoms, the patient reported inflammatory back pain that had been evolving for 2 years and had worsened in recent months.

- Clinical Examination:
  - Pulmonary auscultation: unremarkable
  - Pain on palpation of the spinous processes of D3-D4-D5
  - No neurological deficit
- Radiographic Findings:
  - Chest X-ray **(Figure 1)** revealed an opacity in the right pulmonary apex, measuring 3 cm in its longest dimension, poorly defined, and homogeneous with the upper right lobe. There was also condensation of the T4 vertebra without apparent bone lysis.

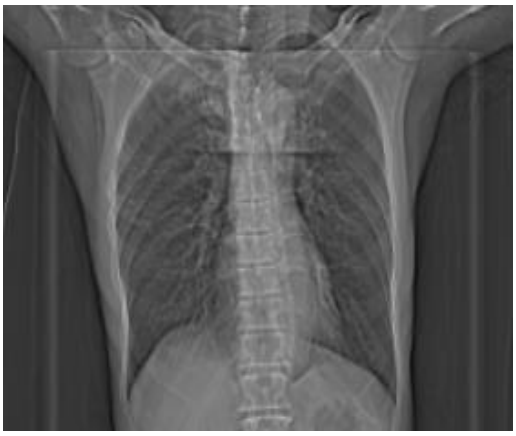

Figure 1: Opacity of the right pulmonary apex 3 cm in long axis, poorly limited in the homogeneous right upper lobe with condensation of the T4 vertebra without probable bone lysis

- Thoraco-abdomino-pelvic CT scan revealed a tumor mass in the apical segment of the right upper lobe invading the thoracic wall **(Figure 2)**, classified as T3N0M0, and a predominantly fatty density mass in the right costo-vertebral groove at the level of T4 with foraminal extension at T4-T5 and postero-lateral intraspinal extension. The vertebral body and the posterior arch of T4 exhibit a heterogeneous appearance consistent with an angiomyolipoma **(Figure 3)**.

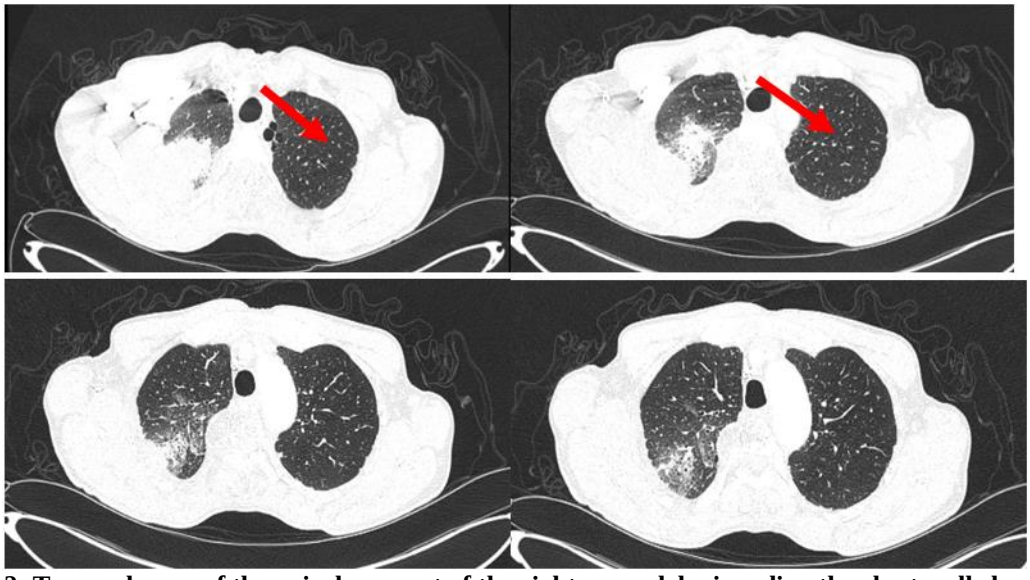

Figure 2: Tumoral mass of the apical segment of the right upper lobe invading the chest wall classified as T3N0M0

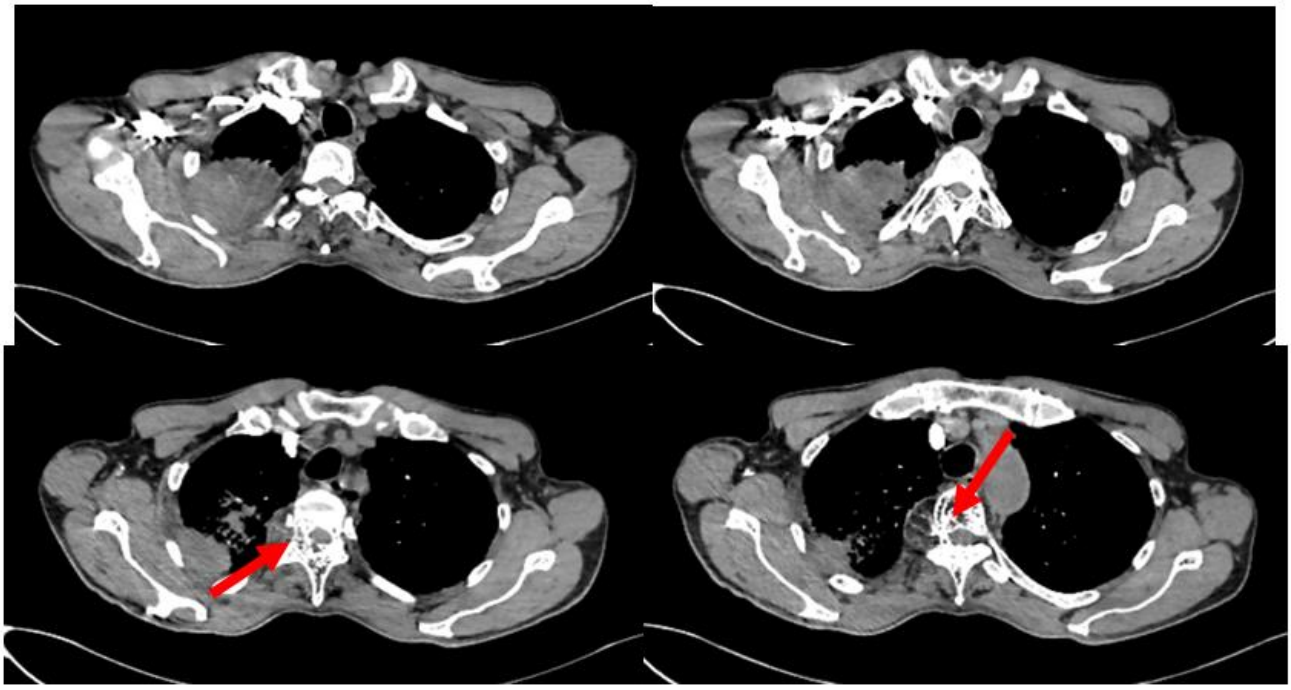

Figure 3: Heterogeneous appearance of the vertebral body and posterior arch of T4 suggesting an angiomyolipoma

MRI showed multiple vertebral angiomas **(Figure 4)**: A nodule in the vertebral bodies of T2 and T3 with isosignal on T1, hypersignal on T2, suggestive of a vertebral hemangioma, and a mass at the T4 vertebra involving the right pedicle of T4 **(Figure 5)**. This mass extends into the right posterior mediastinal space at the costo-vertebral groove and the T4-T5 intervertebral foramen, measuring 38*33 mm, with T1-T2 hypersignal and T1 Fatsat hyposignal, enhancing after gadolinium injection, consistent with an aggressive vertebral angioma (AVA).

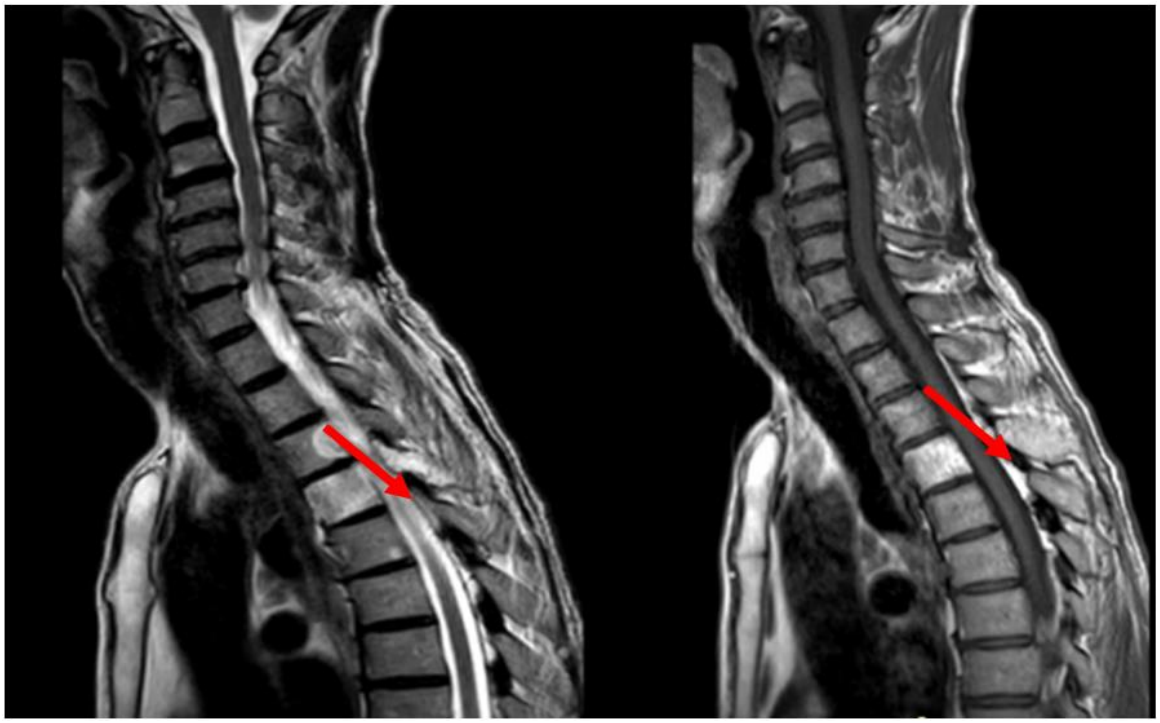

Figure 4: T2 and T3 vertebral body nodule in T1 isosignal, T2 hypersignal suggesting a vertebral angioma

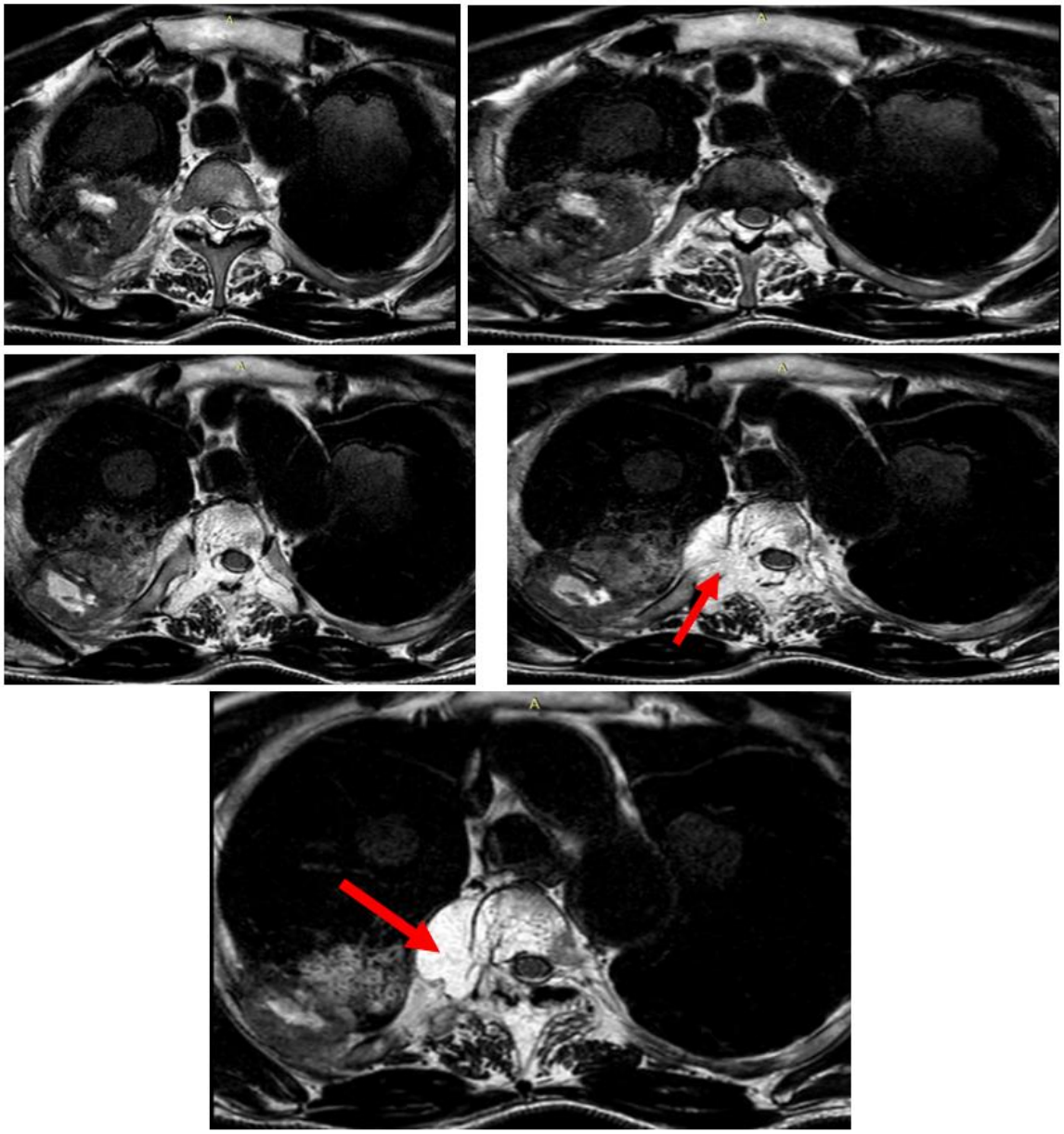

Figure 5: Mass at the level of the T4 vertebra which involves the right pedicle of T4 and extends to the right posterior mediastinal space at the level of the costovertebral groove and at the level of the T4-T5 intervertebral foramen measuring 38*33mm in T1-T2 hypersignal T1 hyposignal Fatsat enhancing after the injection of gadolinium consistent with an aggressive vertebral angioma (AVA)

These benign lesions remained asymptomatic and are discovered incidentally during systematic examinations as part of the staging workup. However, due to the aggressive nature of the D12 angioma and its close contact with the pulmonary tumor in the posterior mediastinum, surgical management was considered.

A multidisciplinary approach was organized, and the indication was to perform preoperative embolization, followed by hemivertebrectomy of D12 associated with costolobectomy.

The patient refused to continue the treatment and passed away after 5 months.

DISCUSSION

Most vertebral angiomas are discovered incidentally and should not be treated [5]. Spinal pain is generally the most common symptom of discovery [6] similar to our reported case. MRI remains the most effective in exploring vertebral angiomas [5], the amount of vascular and adipose tissue could predict the behavior of the lesion [7, 8] Between 0.9 and 1.2% of vertebral angiomas are aggressive [9, 10] and can be extended to the medullary canal or paravertebrally which can lead to a neurological deficit [11, 12], the therapeutic choice of AVA is controversial having a wide therapeutic arsenal including endovascular embolization, radiotherapy, percutaneous sclerotherapy, surgery ranging from vertebroplasty to total spondylectomy, thus various

combinations of these methods have been used. A treatment algorithm has been recommended by some authors who propose radiotherapy alone for small lesions where vertebral stability is not compromised, vertebroplasty associated or not with embolization for small hemangiomas without invasion of the root canal but symptomatic with pain, embolization associated with surgery for large painful AVAs with or without invasion of the spinal canal.

AVAs represent a significant hemorrhagic risk during resection, hence the interest in preoperative endoarterial embolization. Daniel [13] shows in a study conducted on 10 patients operated between 1990 and 2016 for symptomatic AVAs due to pain (two cases) or neurological signs (eight cases) that preoperative embolization followed by tumor resection and spinal stabilization is a safe and effective procedure. In another retrospective study involving 20 patients with invasive vertebral angiomas who received different types of surgery and divided into two groups, group A (11 cases) who received preoperative embolization and group B (9 cases) operated without embolization, Fan and Hao [14] conclude that intraoperative blood loss, transfusion volume, and operating time decrease significantly with preoperative embolization (group A).

In a meta-analysis of 17 studies and publications, Robinson [15] shows that for groups that had undergone preoperative embolization, bleeding was estimated at an average of 960cc compared to 1629cc for non-embolized groups, concluding with a strong recommendation for embolization before surgery. However, this technique is not without complications: allergic reaction to the embolization product, spinal cord ischemia due to accidental embolization of a radiculomedullary artery, bleeding due to early revascularization of the tumor.

The use of surgical wax reduces intraoperative blood loss as well as operating time in surgery for compressive thoracic vertebral hemangiomas [16].

Radiotherapy would also be an effective and safe treatment for AVAs in the event of spinal cord compression or neurological deficit requiring additional surgery [17].

Pancoast-Tobias tumor (PTT) can extend to the cervicothoracic outlet divided into three compartments (ant, middle, post) each of which can be invaded in isolation and be responsible for different symptoms [18,19]. At the time of diagnosis of this type of tumor, thoracic symptoms have already been present for months (5 to 6 months) [18] as in our case presented, and few patients do not have any [1]. Pain is the most common symptom, which can present as chest pain or pain radiating to the shoulder as in our patient's case [18].

The diagnosis of these tumors of the pulmonary apex calls for standard chest X-ray as first-line examination, often showing pulmonary opacity encompassing the apex, with sometimes visible osteolysis of the ribs and spine [19], thoraco-abdomino-pelvic CT scan is part of the extension assessment and MRI allows for precise analysis [20] of the extension of the tumor to the brachial plexus, the spine as well as invasion of the conjugation foramina. The central problem for these tumors remains the selection of candidates for surgery with the essential objective of complete resection. TPTs can be resected by two approaches: posterior approach by posterolateral thoracotomy and anterior approach by cervico-sterno-thoracotomy. The largest single-center series published to date remains that of the Memorial Sloan Ktering Center with 225 patients treated between 1974 and 1988 where a complete resection was performed in 64% of patients with tumors classified as T3N0, with a 5-year survival of 46% in the case of patients classified as T3N0M0.

Moussa and Xavier [21] evaluated the results of a 63-year surgical experience of tumors of the parieto-pulmonary apex that had a surgical intervention with curative intent by en bloc parieto-pulmonary excision either by lobectomy (88.9%), or by atypical resection (11.1%), a vascular extension (n = 4) or spinal (n = 2) was necessary in 6 cases. The rate of complete resection was 84.1% and the factors identified in the analysis as significantly correlated with better survival were the complete resection R0 and the T3N0M0 status as in the case of our patient.

CONCLUSION

Vertebral hemangioma is a benign tumor that can be aggressive and cause serious neurological complications, although no treatment is necessary in the presence of quiescent forms or clinical and radiological monitoring is advised. The selection of TPT with the objective of complete resection often represents a good prognosis at 5 years. The management of the exclusive association of a tumor of the pulmonary apex and aggressive vertebral angiomas remains a a challenge.

Authors contribution

El Ayachi Montassar: original draft writing
Mahdi Meddeb: Data analysis, Paper validation.
Ahmed Mzid: Contributor
Yassine Aloui: Writing paper
Khalil Habboubi: Supervision
Mondher Mestiri: Supervision

Conflict of interest: No

REFERENCES

- Pilegaard, H., & Licht, P. B. (2017). Minimal invasive repair of pectus excavatum and carinatum. *Thoracic Surgery Clinics*, *27*(2), 123-131.

- Murugan, L., Samson, R. S., & Chandy, M. J. (2002). Management of symptomatic vertebral hemangiomas: review of 13 patients. *Neurology India*, *50*(3), 300-305.
- Aich, R. K., Deb, A. R., Banerjee, A., Karim, R., & Gupta, P. (2010). Symptomatic vertebral hemangioma: treatment with radiotherapy. *Journal of cancer research and therapeutics*, *6*(2), 199-203.
- Nassar, S. I., Hanbali, F. S., Haddad, M. C., & Fahl, M. H. (1998). Thoracic vertebral hemangioma with extradural extension and spinal cord compression: Case report. *Clinical imaging*, *22*(1), 65-68.
- Hiari, A., Nawaiseh, B., & Jaber, H. (1998). Magnetic resonance imaging in the diagnosis of vertebral haemangiomas. *EMHJ-Eastern Mediterranean Health Journal, 4 (1), 149-155, 1998*.
- Laredo, J. D., Reizine, D., Bard, M., & Merland, J. J. (1986). Vertebral hemangiomas: radiologic evaluation. *Radiology*, *161*(1), 183-189.
- Hillman, J., & Bynke, O. (1991). Solitary extradural cavernous hemangiomas in the spinal canal: Report of five cases. *Surgical neurology*, *36*(1), 19-24.
- Nguyen, J. P., Djindjian, M., Gaston, A., Gherardi, R., Benhaiem, N., Caron, J. P., & Poirier, J. (1987). Vertebral hemangiomas presenting with neurologic symptoms. *Surgical neurology*, *27*(4), 391-397.
- Slon, V., Stein, D., Cohen, H., Sella-Tunis, T., May, H., & Hershkovitz, I. (2015). Vertebral hemangiomas: their demographical characteristics, location along the spine and position within the vertebral body. *European Spine Journal*, *24*, 2189-2195.
- Heyd, R., Seegenschmiedt, M. H., Rades, D., Winkler, C., Eich, H. T., Bruns, F., ... & German Cooperative Group on Radiotherapy for Benign Diseases. (2010). Radiotherapy for symptomatic vertebral hemangiomas: results of a multicenter study and literature review. *International Journal of Radiation Oncology* Biology* Physics*, *77*(1), 217-225.
- Fox, M. W., & Onofrio, B. M. (1993). The natural history and management of symptomatic and asymptomatic vertebral hemangiomas. *Journal of neurosurgery*, *78*(1), 36-45.
- Kato, S., Kawahara, N., Murakami, H., Demura, S., Yoshioka, K., Okayama, T., ... & Tomita, K. (2010). Surgical management of aggressive vertebral hemangiomas causing spinal cord compression: long-term clinical follow-up of five cases. *Journal of orthopaedic science*, *15*, 350-356.
- Eichberg, D. G., Starke, R. M., & Levi, A. D. (2018). Combined surgical and endovascular approach for treatment of aggressive vertebral haemangiomas. *British journal of neurosurgery*, *32*(4), 381-388.
- Fan, H., Hao, D., Guo, Y., Zhang, X., Gao, W., Wang, X., & Zhu, J. (2019). Effect of preoperative feeding artery occlusion on invasive vertebral hemangioma resection. *Zhongguo xiu fu Chong Jian wai ke za zhi= Zhongguo Xiufu Chongjian Waike Zazhi= Chinese Journal of Reparative and Reconstructive Surgery*, *33*(5), 606-611.
- Robinson, Y., Sheta, R., Salci, K., & Willander, J. (2015). Blood loss in surgery for aggressive vertebral haemangioma with and without embolisation. *Asian spine journal*, *9*(3), 483.
- Cui, X., Li, L. T., & Ma, Y. Z. (2016). Anterior and posterior instrumentation with different debridement and grafting procedures for multi-level contiguous thoracic Spinal tuberculosis. *Orthopaedic surgery*, *8*(4), 454-461.
- Wang, B., Meng, N., Zhuang, H., Han, S., Yang, S., Jiang, L., ... & Liu, Z. (2018). The role of radiotherapy and surgery in the management of aggressive vertebral hemangioma: a retrospective study of 20 patients. *Medical science monitor: international medical journal of experimental and clinical research*, *24*, 6840.
- Strojan, P., Debevec, M., & Kovač, V. (1997). Superior sulcus tumor (SST): management at the Institute of Oncology in Ljubljana, Slovenia, 1981–1994. *Lung cancer*, *17*(2-3), 249-259.
- O'Connell, R. S., McLoud, T. C., & Wilkins, E. W. (1983). Superior sulcus tumor: radiographic diagnosis and workup. *American Journal of Roentgenology*, *140*(1), 25-30.
- Panagopoulos, N., Leivaditis, V., Koletsis, E., Prokakis, C., Alexopoulos, P., Baltayiannis, N., ... & Dougenis, D. (2014). Pancoast tumors: characteristics and preoperative assessment. *Journal of thoracic disease*, *6*(Suppl 1), S108.
- Ouattara, M., D'Journo, X., Avaro, J. P., Trousse, D., Loundou, A., Doddoli, C., ... & Thomas, P. Tumeurs de l'apex pariéto-pulmonaire: une expérience chirurgicale monocentrique de 63 cas.

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