

Solitary Fibrous Tumor and Parathyroid Adenoma: Case Report

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ARTICLE INFO

Article history:

Received: 18 August 2018;

Received in revised form:
20 September 2018;

Accepted: 1 October 2018;

Keywords

Association,
Solitary Fibrous Tumor,
Mediastinum,
Parathyroid Adenoma,
Surgery,
Immunohistochemistry.

ABSTRACT

Solitary fibrous tumor (TFS) is a rare tumor. It is localized preferentially at the pleura. Other locations have been described. These are peritoneal, bronchopulmonary, orbital and mediastinal locations. We report the case of a 49-year-old patient followed for parathyroid adenoma who presented with bone pain, muscle pain, fatigue, dry cough and dyspnea. The clinical examination was without abnormality. Chest X-ray showed a superior mediastinal opacity. Cervicothoracic computed tomography indicated a mass at the lower right pole of the thyroid and a tissue process of the upper mediastinum. A cervicotomy was performed for the removal of the parathyroid adenoma. On the other hand, an attempt at dissection of the mediastinal mass by the cervical route was inconclusive, and a manubriotomy was necessary for its extraction. The histopathological study confirmed the parathyroid adenoma for the cervical mass; and solitary fibrous tumor for the mass of the superior mediastinum.

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INTRODUCTION

Solitary fibrous tumors are characterized by their scarcity and benignity and are often localized in the pleura. They occur at any age especially between the 5th and 7th decade [1]. They are asymptomatic and of accidental radiological discovery. Several localizations have been described, including mediastinal localization. Its diagnosis is confirmed by surgery and the histology associated with the immunohistochemical study. They are often of mesenchymal origin and may recur.

CASE REPORT

Mrs. F, 49 years old, with a complex medical history of type 2 Diabetes for 5 years under oral antidiabetics; an autoimmune hypothyroidism under LEVOTHYROX;

dyslipidemia since 2011 under CRESTOR; a benign intracranial hypertension under LAROXYL since 2015; was operated on in 2015 for a fusing and arachnoidocele carpelitis and for a synovial cyst in 2009; and currently being followed for a parathyroid adenoma since 2015. She presented with bone and muscle pain, fatigue, abdominal pain, constipation, anorexia, anxiety and impaired concentration, a dry cough and mild progressive dyspnea for 4 months. A chest X-ray showed opacity at the tissue level. A cervicothoracic computed tomography showed a cervical mass behind the lower pole of the right lobe of the thyroid and another tissue process in the upper mediastinum. Biologically, her parathyroid hormone was measured to 157.6 µg / ml and her serum calcium was 103.

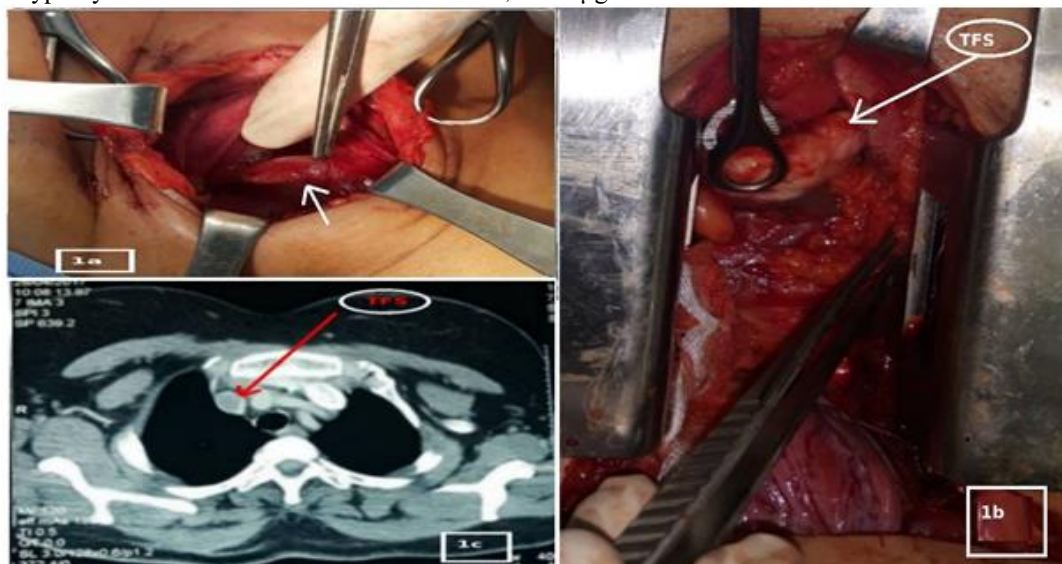


Fig 1a. Parathyroid adenoma intraoperatively.

Fig 1b. Solitary fibrous tumor intraoperatively.

Fig 1c. chest CT scan showing solitary fibrous tumor.

An intervention was necessary for the removal of the parathyroid adenoma and the mass of the superior mediastinum. A cervicotomy was performed allowing resection of the parathyroid adenoma (fig 1a). An attempt at dissection of the mediastinal mass by cervical surgery failed. A manubriotomy was necessary for its extraction (fig. 1b). The postoperative procedure was without complication. The anatomopathological study of operative specimens returned to favor a parathyroid adenoma for the cervical nodule and a solitary fibrous tumor for the anterior mediastinal mass. In the immunohistochemical study of the mediastinal mass, the tumor cells indicated CD34 with a low and focal immunolabeling. The antibodies PS100, B CATENINE and AML were all negative.

DISCUSSION

Solitary fibrous tumors of the pleura were described for the first time in 1931 by Klemperer and Rabin [1]. It is a heterogeneous group of benign and malignant tumors of mesenchymal origin [2] located in the majority of cases in the pleura. Mediastinal localization is rare and aggressive and represents 1-8% [3]; it was first described in Japan in 1992 by Fukushima [4]. These tumors are generally benign but can evolve towards malignancy in 13% [5].

These tumors can occur at any age with a predominance between 50 and 70 years without predominance of sex [6]. According to a study by Witkin et al [7], the age range of patients were 27 to 70 years with an average age of 54 years. On the clinical level, solitary fibrous tumors are asymptomatic and sometimes expressed by chest pain, cough, dyspnea and very rarely by hemoptysis or obstructive pneumonia [5]. They can be expressed by signs of compression that depend on the seat and the volume of the tumor, namely dysphagia, an upper vena cava syndrome or disorders of the heart rhythm [4]. These tumors can in some cases be revealed by extrathoracic manifestations [10], namely:

- The hypertrophic osteoarthropathy of Pierre Marie found in 16% of cases,
 - Hypoglycemia attacks found in 1.3-4% in relation to tumor secretion of insulin-like substance type II [5] but whose mechanism remains unknown,
 - A digital clubbing probably related to an abnormal production of hepatocyte growth factor (IGF).
- None of these signs were found in our patient.

The means of diagnosis of these tumors are various but only the histology allows the definitive diagnosis. On chest X-rays, they appear as a well-defined, homogeneous, lobulated opacity with clear external boundaries and a gently sloping connection to the mediastinum and internal limits embedded in the mediastinum [4]. CT is also essential. It is useful in confirming the tissue nature of the mass and it also specifies the limits of the mass with neighboring organs. They generally appear on CT as homogeneous and well-circumscribed masses or in the form of heterogeneous masses with zones of necrosis, calcification and cystic or myxoid degeneration [9], which constitute factors of poor prognosis. However, it is difficult to differentiate at this stage this tumor with other mediastinal tumors i.e. thymic tumors, peripheral nerve tumors, sarcomatoid carcinomas, lymphomas, metastases, various sarcomas, myofibroblastic inflammatory tumors [6]. For our patient, the radiological presentation was misleading because it suggested a thymic tumor initially before the histology confirmed that it was a solitary fibrous tumor of the mediastinum. MRI is essential especially when vascular invasion of the mass is suspected and allows a three-

dimensional analysis of the mass [10]. MRI and pet-scan can also be useful in particular by distinguishing a benign tumor from a malignant tumor by detecting possible metastases [3]. Angiography can also be used for the diagnosis of these tumors because of its very high vascular nature [3]. Although these different examinations may be useful in the diagnosis of these tumors, a tissue biopsy of the mass is essential in order to determine the definitive diagnosis. Several techniques are used including a needle aspiration biopsy of the tumor [2,3,4]. Nevertheless, this biopsy has certain limitations such as not visualize all of the morphological zones of the tumor and the fragment may return negative.

Histologically, a more or less dense proliferation of fusiform cells is found evoking fibroblasts [2]. A high mitotic index (greater than four mitoses per ten high-magnification fields), high cell density, necrotic-hemorrhagic changes, marked nuclear pleomorphism, and stromal or vascular invasion generally constitute major signs of malignancy [5, 7, 11]. The immunohistochemical study of these tumors is important in the diagnosis of these tumors, their cells often express vimentin, CD99, bcl2, occasionally desmin and negative for S100 and cytokeratin. CD34 is expressed in 90% of cases [2, 4, 5, 12]. In our patient CD34 was positive and PS 100 antibodies were negative. According to a recent study, solitary fibrous tumors have an NAB2-STAT6 and nuclear STAT6 fusion gene, which is a significant advance in the diagnosis of these tumors [2, 3].

The treatment of these tumors is the complete surgical excision widened to neighboring organs affected in order to avoid a possible recurrence likely to cause a bad prognosis. Radiation therapy and chemotherapy can be used mainly in case of incomplete resection of the tumor [2,3,11]. The prognostic criteria for solitary fibrous tumors of the pleura are identical to those of the mediastinum. After complete surgical resection of these tumors the evolution is generally towards the complete remission however in some cases the evolution can be unpredictable or a long-term follow-up is necessary because these tumors evolve in 13 to 23% towards the malignancy [4,5,6]. The association of a solitary fibrous tumor with a parathyroid adenoma is exceptional and has never described in literature to our knowledge.

CONCLUSION

Solitary fibrous tumors of the mediastinum are rare. Complete surgical excision is the basis of treatment which is both diagnostic and therapeutic. Their diagnosis is difficult due of the absence of specific clinical signs with only histology being able to confirm it. The prognosis of these tumors is favorable after surgery; however, a long-term follow-up is necessary to detect a possible recurrence.

REFERENCES

- 1-E. Gomez, B. Marie, S. Tissier, C. Thivillier1, J.M. Vignaud, D. Regent, F. Chabot Une Cause Exceptionnelle De Tumeur Trachéale:La Tumeur Fibreuse SolitaireRev Mal Respir 2007; 24: 81-5 Doi:10.1019/20064208
- 2-Alaina J. Webb, Ahmed S. Yassin, Ali Saeed, Hemang Yadav, and James P. Utz Mediastinal Solitary Fibrous Tumor Diagnosed by Endobronchial Ultrasound-Directed Biopsy Am J Case Rep. 2017;18:549-552.
- 3-Jan De Raet, MD, Robert Sacré, MD, Anne Hoorens, MD, PhD,Christopher Fletcher, MD, FRC Path, and Jan Lamote, MD, PhD Malignant Giant Solitary Fibrous Tumor of the Mediastinum J Thorac Oncol. 2008;3: 1068-1070
- 4-M. Nejjar, K. Hidraoui, W. Mhridi, M. Ridai, N.O. Zerouali, H. El Attar, S. Azzouzi, A. IraqiTumeur Fibreuse Solitaire Du Médiastin: À Propos D'un Cas Rev. Pneumol.

Clin., 2004, 60, 4-235-238

5-F. Chermiti Ben Abdallah *, S. Bousnina, H.Racil, L. Ismail, F. El Mezni, M.L. Megdiche, A. Chabbou Les Tumeurs Fibreuses Solitaires Malignes De La Plèvre Revue de Pneumologie clinique (2010) 66, 187—190

6-Brian R Gannon, Carolyn D O'Hara, Kenneth Reid , and Phillip A IsotaloSolitary Fibrous Tumor Of The Anterior Mediastinum: A Rare Extrapleural Neoplasm Tumori,93: 508-510,2007

7-Witkin GB, Rosai J: Solitary fibrous tumor of the mediastinum. A report of 14 cases. Am J Surg Pathol, 13: 547-557,1989

8-H. Kabiri, L. Achachi, N. Mahassini, S. Al Aziz, A. El Maslout, F. Mansouri, A. Benosman Le Mesotheliome Fibreux De La Plevre: A Propos D'une Observation

Médecine du Maghreb 1999 n°77

9- Hsiou –chun Lin, Yung-Cheng Wang, Hsio-yun Chang, Solitary Fibrous Tumor Of Right Anterior Mediastinum: A Case Report Fu-Jen Journal of Medicine Vol.15 No.1 2017

10- L. Cannard, L. Debelle, V. Laurent, S. Béot, J-C Leclerc, D. Régent imagerie des tumeurs fibreuses de la plèvre feuillets de Radiologie, 2001, 41, n°4, 317-324

11-Hanau CA, Miettinen M. Solitary fibrous tumor: histological and immunohistochemical spectrum of benign and malignant variants presenting at different sites. Human Pathology. 1995;26(4):440-9.

12- Niaz Hussain Soomro, SalwaPervaiz , Ammad Hussain, Syed Khalid Ali Solitary Fibrous Tumor of the Mediastinum: A Rare Tumor at a Rare Site J PIONEER Med, Sci 2016 ; 6(3) : 100-103