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Case report

Obstructive tracheal neoplasm: Primary tracheobronchial non-hodgkin lymphoma

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ABSTRACT

The trachea is an unusual site of primary malignancy. Very few cases of primary tracheal lymphoma with central airway obstruction have been reported so far. Common complaints are dyspnea and cough that could mimic a partially refractory asthma in some cases. In this article; we will present the case of a 63-year-old woman diagnosed with a tracheal lymphoma causing life-threatening airway obstruction, this was confirmed by bronchoscopy biopsy and histopathological exmination.

The mortality depends on the progression of the disease, the obstruction of the airway. However, this entity has a good prognosis if diagnosed immediately and treated with specific chemotherapy.

This case will show that the diagnosis of tracheal lymphoma should be kept in mind within the differential diagnosis of central airway obstruction.

1. Introduction

The primary neoplasm of the trachea is rare [1]., it respresents only 2% of all malignancies [2]. The most common tracheal tumor is squamous cell carcinomas followed by adenoid cystic carcinomas [3].

The tracheal lymphoma is a very rare presentation as it represents less than 3% of all tracheal tumors [4]. The non-specific nature of the symptoms as well as the extremely rare frequency of this entity, always leads to an erroneous diagnosis and to a delayed treatment that could be lethal.

In this article, we report a case of a tracheal lymphoma arising in the distal trachea immediately above the carina. We discuss the imaging caracteristics of this pahology in order to consider tracheal lymphoma among the tracheal tumors and in the differential diagnosis of central airway obstruction.

2. Patient and methods

The patient was a 63-year-old woman,non smoker, with 09 months history of dyspnea, wheezing, cough without chest pain or hemoptysis. The initial diagnosis performed was asthma. However, no improvement has been noted under the asthma treatment. She was admitted in the emergency department for dyspnea (Class III according to the New York Heart Association classification). She was afebrile (Temperature: 36,7 °C), respiration rate (28 cpm); pulse (95 bpm); blood pressure (135/80 mmHg). The physical examination revealed respiratory distress with inspiratory and expiratory stridor. The remaining examination was inconspicuous. The patient underwent urgently nasal continuous positive airway pressure (CPAP). Chest contrast-enhanced computed tomography (CT) with multi-planar reconstruction was done in emergency and showed an irregular, asymmetrical, broad-based nodular circumferential thickness of the anterior and posterior walls of the distal trachea, partially filling the left bronchi, with moderate and homogenous enhancement. The lesion was causing almost total obstruction of the lumen. The CT revealed no other tracheobronchial lesion or extrinsic compression including any mediastinal or hilar lymphadenopathy (Fig. 1). An endoscopic examination of the trachea (Fig. 2) with biopsy were done. Histopathology revealed the marginal zone B-cell lymphoma

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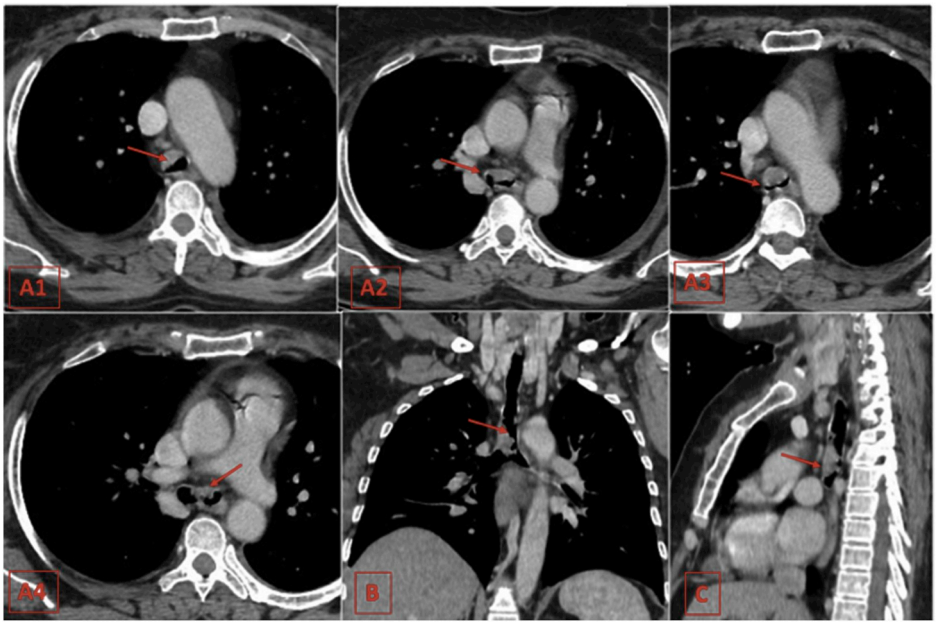

Fig. 1. Axial contrast-enhanced chest computed tomography in (A1,A2,A3,A4) with coronal and sagittal reconstructions in (B) and (C), respectively.
The images are showing an irregular, asymmetrical, circumferential thickness of the anterior and posterior walls of the distal trachea, partially filling the left bronchi, with moderate and homogenous enhancement. This tumor configuration caused almost total obstruction of the lumen.

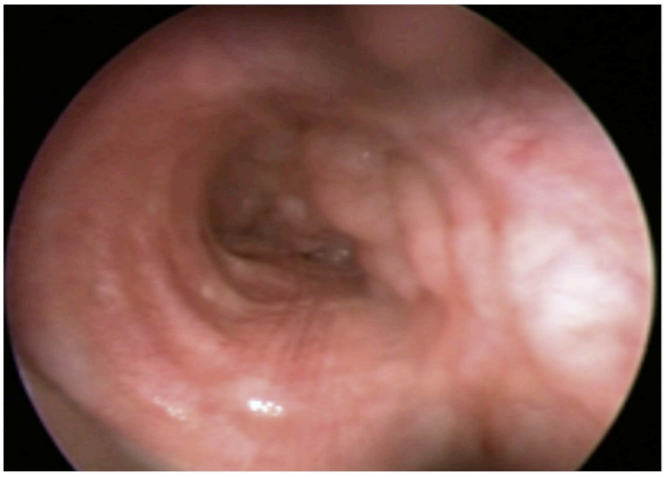

Fig. 2. Bronchoscopy view before intervention showing a budding formation obstructing the distal trachea.

(NHL) positive for CD20 and negative for CD30, CD5, CK19, CD10, Cyclin D1, chromogranin and synaptophysin (Fig. 3). An Y-shaped prosthesis has been installed with satisfactory endoscopic control. The patient underwent chemotherapy (RCHOP) and radiotherapy (16 sessions with a total dose of 30 Gy). The evolution was good with regression of symptomatology especially of dyspnea. Her control CT revealed a regression of the tumor mass (Fig. 4). The patient showed a total improvement of symptoms and no incidents were reported.

3. Discussion

For this study, we used a pubmed research (https://www.ncbi.nlm.nih.gov/pubmed) which allowed us to collect articles as well as references mentioned on available documents.

Even if extranodal lymphoma is common (gastrointestinal tract, cervical region), a primary presentation of extranodal lymphoma involving the trachea is extremely unusual. Primary lymphoma in the trachea originates from B and T epithelial cells that participate in upper airway immunomonitoring.Primary tracheobronchial NHL occurs in less than 1% of all NHL patients [5].

It affects a wide range of individuals in the age category 4–80 years;

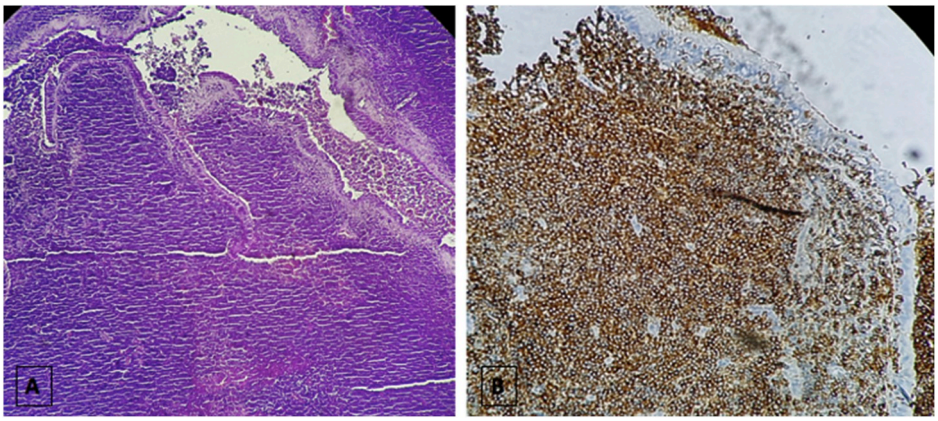

Fig. 3. A: (HESX100) Tumoral proliferation made of small hyperchromatic cells. **B:** Intense and diffuse immunomarking of the tumoral cells by CD20.

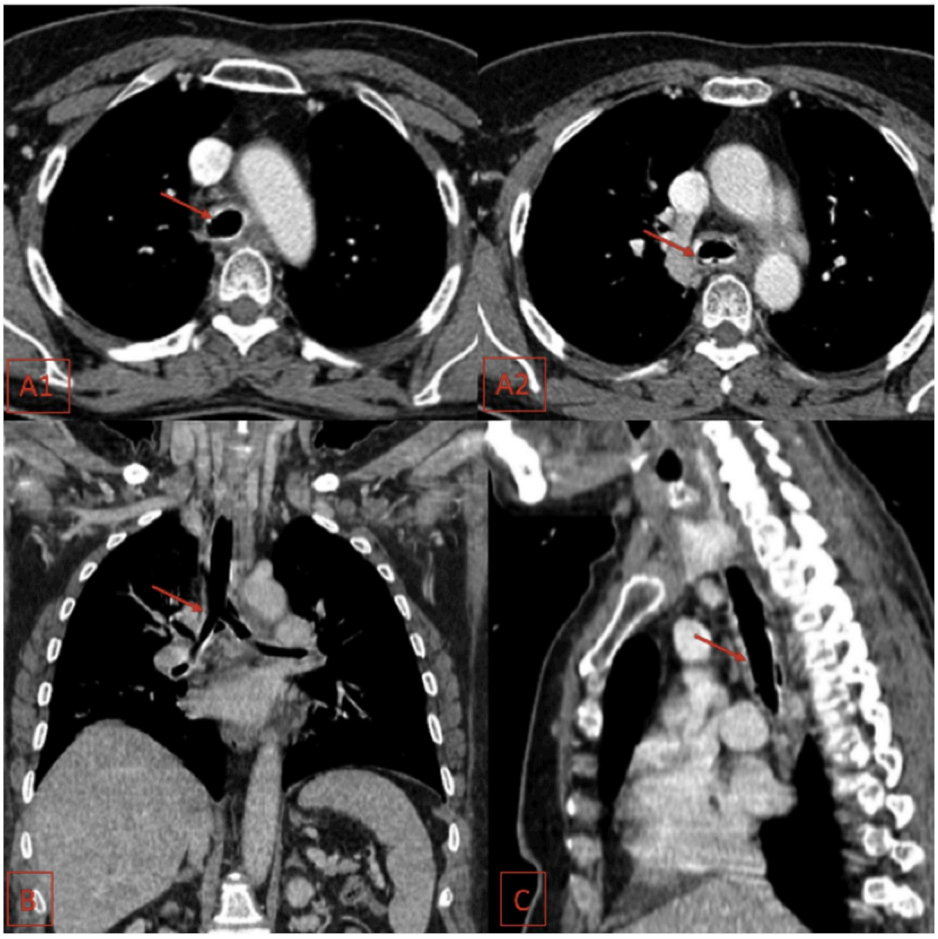

Fig. 4. Axial contrast-enhanced chest computed tomography in (A1,A2) with coronal and sagittal reconstructions in (B) and (C), respectively. The images are showing a total regression of the tumor mass with an Y prothesis in place.

the average age at diagnosis is 45 years [4]. Both males and females can be affected. It can occur worldwide and all races and ethnic groups may be affected.

No specific risk factors have been identified for Lymphoma of Trachea. However, the condition is known to be associated with the following factors: Autoimmune disorders, family history of immune disease, systemic disease, advanced age, smoking, exposure to radiation and industrial chemicals,infection (Viruses in some rare cases, Epstein-Barr virus infection).

The most common symptoms of tracheal lymphoma are non specific and are those of upper airway obstruction: dyspnea, cough, wheezing, stridor. Hemoptysis is uncommon [4].

Since 1973, Thirty-four cases of primary tracheal lymphoma were identified, including our patient (Table 1).

The ages of patients range from 4 to 81 years with a median age of 35 years. The most common symptoms by frequency were dyspnea (58%), cough (53%), wheezing or stridor (38,2%), and hoarseness (11,8%). Only one patient presented hemoptysis (2,9%). Five patients (14,7%) were initially misdiagnosed as bronchial asthma. Median duration of symptoms prior to diagnosis was 1,4 months. It seems to be related to the histological subtypes. Tracheal obstruction developed in 28 (82,3%), including 21 (61,7%) of cases who required stenting or emergency surgery.

Radiological investigations are important for diagnosis. Chest X-Ray is often performed on a first-line, however, tumors may be undetected and the diagnosis is usually delayed or misinterpreted as asthma. The diagnosis may also be delayed in adults due to the large functional reserve of tracheal lumen, symptoms appear after significant occlusion (50–75% of lumen) [4,6].

CT is the most effective imaging modality for detecting tracheal tumors, assessing their extensions, and analyzing the perilesional environment [6]. The CT has crucial role since being sensitive to up to 90% of cases. It was useful to identify the patient's obstructive symptoms source.

Bronchospy is useful for the diagnosis and staging of tumors, as well as obtaining lesion samples.

Various histological subtypes of primary tracheal lymphoma exist. The Hodgkin's lymphoma was the most frequent (06 cases). The second most common subtype was by the mucosa-associated lymphoid tissue (MALT) lymphoma, comprising 5 (14,7%), followed by the anaplastic large cell lymphoma (04 cases) and unspecified T-cell lymphoma (04 cases). In our case, the histopathological examination objectived a marginal zone B lymphoma. The others B cells lymphoma subtypes identified in the study were: diffuse large B cell (n = 3), medium large B cell (n = 1), B imuunoblastic (n = 1), polymorphic B cell (n = 1).

The median age for diagnosis of marginal zone lymphoma is 65 years old, in our case the patient has 63 yeras old.

Marginal zone lymphomas (MZL) are slow growing and make up about 12% of all B-cell NHL [37].

MZL are considered low grade B cell NHL. They are developed as their name suggest in the marginal zone of lymphoïd tissue [38]. There are 3 categories of MZL: Splenic, nodal and extra nodal. They represents both for the nodal and splenic less than 0,02% and for tyhe MALT 0,13% [39,40].

Marginal zone lymphomas generally lack markers in order to come up with an overall diagnosis. CD20 antibodies were found to be present

Table 1
Characteristics of primary tracheal lymphoma patients.

No	Year of diagnosis	Age, y/sex	Symptoms	Duration of symptoms prior to diagnosis	Tracheal stenosis	Histology	Treatment	Outcome	Reference
1	ND	ND	ND	ND	ND	Lymphocytic	R	Died,>1 y	[9]
2	1973	81/W	Dyspnea	1 month	Y	Diffuse well-differentiated	S, R	CCR,>22 mo	[10]
3	1975	67/M	Dyspnea, wheeying, cough, misdiagnosis as asthma	9 months	Y	Mixed type	S	CCR,>64 mo	[11]
4	ND	28/M	Dyspnea, cough, fever	3 months	Y	Hodgkins	C, R	CCR,>8 mo	[12]
5	1983	65/F	Dyspnea, wheezing, stridor	7 months	Y	Diffuse lymphoplasmacytic	S, R	CCR,>22 mo	[14]
6	ND	63/M	Wheezing, cough, misdiagnosis as asthma	6 months	Y	MALT, B-cell	R	CCR	[15]
7	1983	17/M	Hoarseness, cough, chest pain	ND	Y	Diffuse large histiocytic	R, C	Died, early	[16]
8	ND	17/M	Superior vena cava syndrome	ND	Y	Lymphoblastic	S, C, R	Died, early	[16]
9	ND	46/M	Stridor, cough, hoarseness, shortness of breath	1 month	Y	T-cell	R, C	CCR,>7 y	[17]
10	1985	74/F	Dyspnea, wheezing	1 y	Y	B- immunoblastic lymphona	S, C	No reccurrence	[13]
11	ND	16/F	Stridor, sore throat, fever,fatigue hemorrhage	4 months	Y	Polymorphic B-cell	Interferon, y-globulin, S, stending	CCR	[18]
12	ND	66/F	Shortness of breath, stridor, cough	2 months	Y	MALT, B-cell	S, R	CCR, >12 mo	[19]
13	ND	44/F	Dyspnea, hoarsenes	Sudden onset	Y	Diffuse medium-sized cell, B-cell	S, C, R	CCR, >12 mo	[21]
14	1987	59/M	Dyspnea, wheezing, cough	Sudden onset	Y	Lymphocytic	R	CCR, >41 mo	[20]
15	ND	4/M	Dyspnea, cough	2 weeks	Y	T-cell	C	ND	[22]
16	1990	9/M	Dyspnea, cough	1 month	Y	T-cell	No	Died, early	[22]
17	1988	30/M	Dyspnea, cough, dysphonia, fever	3 days	Y	Diffuse large cell immunoblastic	C, R	Died, early	[23]
18	ND	52/M	Dyspnea, wheezing, cough, misdiagnosis as asthma	? Several days	Y	Anaplastic large cell	C, R, S	CCR, >13 mo	[24]
19	1998	60/M	Cough, fever	4 months	N	Anaplastic large cell	L, C	CCR, died, 6 mo	[27]
20	ND	64/F	Dyspnea	ND	Y	Hodgkin, nodular-sclerosing type	Stenting, C, R	Alive	[25]
21	ND	31/F	Dyspnea	ND	Y	Hodgkin, nodular-sclerosing type	Stenting, C, R	Alive	[25]
22	1996	16/F	Dyspnea	ND	Y	Large cell, B-cell	Stenting, C, R	Alive	[25]
23	ND	74/F	N	0	N	MALT	L	CCR, >12 mo	[28]
24	ND	49/M	Dyspnea, wheezing, cough	Sudden onset	Y	Lymphoplasmacytoid	S, C	CCR, >12 mo	[26]
25	ND	67/M	Cough	ND	N	MALT	C	CCR, died, 11 mo	[29]
26	ND	16/M	Dyspnea, cough, stridor, misdiagnosis as asthma	2 weeks	Y	Anaplastic plasmacytoma or plasmablastic lymphoma (undetermined)	Stenting, L, C	CCR, >12 mo	[30]
27	ND	44/F	Dyspnea	ND	Y	MALT	S	CCR, >53 mo	[33]
28	ND	35/F	Cough, sputum, dyspnea	2 months	Y	T-cell	C	Died, 1 mo	[32]
29	2000	20/F	Dyspnea, hoarseness	1 day	Y	Anaplastic large cell	S, C, R	CCR, >60 mo	[4]
30	2004	23/F	_	3 years	Y	Hodgkin	S	Relief of symptoms	[31]
31	2011	35/M	Dry cough, fever	1 year	Y	Hodgkin	S	Relief of symptoms	[34]
32	2012	54/F	_	4 years	Y	Hodgkin	S,C	Relief of symptoms	[35]
33	2014	57/F	Stridor, dyspnea, cough, hemoptysis	6 months	Y	Diffuse large B cell lymphoma	S,C, R	Rapid regrowth of tumor, tracheotomized	[36]
34	2019	63/F	Dyspnea, wheezing, cough,misdiagnosis as asthma	9 months	y	Marginal zone B Lymphoma	C, R	Relief of symptoms	current study

*ND indicates not documented; F, Female; M, male; Y, yes;; N, no; S, surgery; R, radiation; C, chemotherapy; L, laser therapy; CCR, continuous complete remission; MALT, mucosa-associated lymphoid tissue.

in 100% of the cases [41].

The samples in our case were positive only for CD20.

The treatment stratgies of tracheal lymphoma are variable. In some cases, radiotherapy or surgery alone or in combination have been suggested [7]. For patients with symptomatic tracheal stenosis, a temporary stent may be initially performed followed by chemotherapy, radiotherapy, or both [8]. primary tracheal lymphomas are usually sensitive to both chemotherapy and radiotherapy.

4. Conclusion

Tracheal lymphoma is a rare tumor and non-Hodgkin's type of lymphoma is a very rare presentation. Despite its rarity, it can have acute presentation because of compromised conduit access to the lungs. It might requires an immediate surgical intervention. Clinicians should be aware of this possibility.Diagnosis was confirmed by biopsy and histopathologic examination and had a favorable response to chemotherapy and radiotherapy.

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Declaration of competing interest

This manuscript has not been published and is not under consideration for publication elsewhere. We have no conflicts of interest to disclose.

Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.rmcr.2020.100995.

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