ISSN No:-2456-2165

Secondary Chylothorax to Substernal Goiter: Case Report and Review of the Literature

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Abstract:- Substernal goiters are frequently associated with symptoms of local compression, including dyspnea, dysphagia and hoarseness. Rarely, an enlarged thyroid can cause venous compression of the mediastinal structures, compression of the thoracic duct is exceptional and may be manifested by chylothorax.

We report a rare case of a 36-year-old woman with compression of the thoracic duct by a large goiter who presented with dyspnea. After undergoing chest drainage associated with a low-fat oral diet, the patient was treated with thyroidectomy.

A trans-cervical thyroidectomy with ligation of the thoracic duct was performed without the need for a sternotomy. This led to resolution of her symptoms. Confirmation of resolution of chylothorax was obtained by postoperative CT scan. Chylothorax is a rare sequel to substernal goiter. It can be managed by thyroidectomy, the trans-cervical approach remains feasible. Sternotomy was avoided in this case

Keywords:- *Sub-Sternal Goiter- Chylothorax- Surgery.*

I. INTRODUCTION

Substernal goiter or plunging goiter represents 7% of mediastinal masses, it is part of the benign pathology of the mediastinum and represents 2% to 15% of all goiter, Designated for surgery (1).

The clinical presentation is very varied, can range from an asymptomatic neck mass to symptoms related to compression of adjacent structures, including dyspnea, dysphagia and hoarseness.

Compression goiter usually causes narrowing of the trachea and compression of the esophagus or jugular vein and narrowing of the esophagus. Rarely, it can be responsible for venous compression of the mediastinal structures.

Compression symptoms are a common indication for thyroidectomy. Chylothorax due to compressive goiter remains a very rare and exceptional complication Due to anatomical relationships, chylothorax on the right side is caused by a very large and low goiter. We report the rare case of chylothorax caused by a large substernal thyroid goiter compressing the thoracic duct, Chylous pleural effusion was treated by Trans-cervical thyroidectomy with ligation of the thoracic duct.

II. OBSERVATION

This is a 36-year-old patient with asymptomatic multi-nodular goiter for 4 years, who presented with a respiratory distress with stage III dyspnea of the MMRC and a dry cough, progressing in a context of apyrexia and deterioration of the general condition, The clinical examination revealed a conscious patient, afebrile, polypneic with an oxygen saturation at 89% and the presence of a low anterior cervical swelling, and a bilateral fluid effusion syndrome at the pleuropulmonary examination.

The chest x-ray [Figure 1], showed the presence of bilateral pleurisy predominantly on the right, a pleural puncture was performed on the right side bringing a fluid of lactescent appearance exudative to 80% with LDH levels and TG in pleural fluid raised, suggesting chylothorax.

As part of the etiological assessment, a thoracic CT [Figure2,3] was performed revealing a compressive goiter at the expense of the left lobe with the presence of bilateral pleural effusion (Secondary chylothorax to compression of the thoracic duct by a plunging goiter was retained), it was completed by a cervical ultrasound which showed a goiter classified eu-TIRADS III. The patient benefited from right chest drainage with an oral diet low in fat, high in protein and in medium-chain TG.

Given the persistence of the chylous effusion despite the thoracic drainage and the prescribed diet, a total thyroidectomy with cervical lymph node dissection was performed, by approach Trans-cervical, A sternotomy was not necessary for the removal, the evolution was marked by the persistence of chylothorax despite the removal of the goiter requiring surgical revision for ligation of the thoracic duct, intraoperatively no leakage was detected.

ISSN No:-2456-2165

After 48 hours, an improvement in respiratory symptoms was noted with disappearance of the chylothorax one week later, the pathological examination of the operative specimen concluded with an encapsulated papillary oncocytic carcinoma of the right lobe of the thyroid measuring 1.4 cm without vascular emboli. The evolution was marked by the disappearance of chylothorax with improvement of the patient's respiratory function.

III. DISCUSSION

The Chylothorax is defined by the presence of chyle in the pleural space and usually results from a disturbance or obstruction of the thoracic duct [2]. The causes of chylothorax are multiple: it can be spontaneous or traumatic (iatrogenic), sometimes congenital. It is also common after thyroid surgery [3] and in association with Graves disease [4]. It has also been described in association with thyrotoxic cardiomyopathy.

The Chylothorax due to substernal goiter is an exceptional manifestation; it is rarely described with only four cases reported in the English literature. [5] A pleural puncture of the fluid with measurement of the content of triglycerides is a key examination, a concentration above 110 mg / l, strongly supports the diagnosis.

Substernal goiter is a special feature in thyroid surgery. There is no standard definition of it. Some authors define substernal goiter as an extension of the thyroid gland below the thoracic inlet by more than 50%, while others include all patients with at least part of the thyroid gland beyond the thoracic entry.

The anatomical and topographical conditions, the size and the nature of the goiter are factors which explain the diversity of the circumstances of discovery of the sub-sternal goiter.

Substernal goiters can remain asymptomatic for a long time, they can be discovered incidentally during a chest x-ray. However, goiter generally progresses to a gradual growth which gives signs of compression of the trachea, esophagus, large vessels and nerves, thus the most frequent clinical symptomatology is exertional dyspnea associated with Stridor, wheezing at rest when tracheal compression becomes severe (luminal diameter less than 5 mm). Other symptoms may be present such as dysphagia, obstructive sleep apnea, cough, superior vena cava syndrome, hoarseness or recurrent nerve palsy.

The suspected mechanism for the onset of chylothorax is external compression of the thoracic duct by a submerging goiter. The thoracic duct is the largest lymphatic duct in the body, responsible for lymphatic drainage of the whole body except for the right sides of the head and neck, it originates between L2 and D11 from the meeting of the two right lumbar collectors and left the lymphatic trunks and the intestinal trunk, it extends along the right side of the esophagus before crossing to the left side in the mediastinum at the level of the fifth or sixth thoracic vertebra, It then travels up the along the esophagus before turning the left and draining the left internal jugular vein and the subclavian vein. Compression can occur anytime along its path. A lower goiter can cause compression if it has significant posterior extension, and especially if it extends down into the thoracic entrance.

This is the case with our patient, who presented with a large compressive goiter at the expense of the left lobe with significant extension both posterior and inferior.

External compression of the thoracic duct by a substernal goiter has rarely been described as a cause of chylous pleural effusion

Of the four cases described in the literature, treatment required sternotomy and even thoracic duct repair in one case [5].

Regardless of chylothorax, the decision on the optimal treatment of compressive goiter in a symptomatic patient remains a challenge. Surgery remains the standard treatment when symptoms of compression are present; yet surgery for substernal goiter is associated with high rates of complications [6]

There are many major complications, namely: paralysis of the recurrent nerve of the larynx (if it is bilateral) with bilateral paralysis of the vocal cords, hypoparathyroidism (when the goiter is extensive and the anatomical landmarks are obscured) and tracheomalacia (due to destruction of the tracheal rings by goiter).

External compression can be relieved with the removal of the goiter without resorting to thoracic duct repair. This should be determined at the time of surgery depending on whether the leak has been identified or not. If the mechanism of chylothorax is by external compression, then it is unlikely that a chyle leak will be identified in the neck. However, if a leak is identified at the time of surgery, it can be repaired. This repair can be done by a trans-cervical or mediastinal approach depending on the location of the leak. As a general rule, this involves the ligation of the thoracic duct, in our patient's case no leakage of the thoracic duct was detected intraoperatively, on the other hand a ligation of the thoracic duct was performed when the chylothorax persisted despite a total thyroidectomy.

This case highlights this rare complication and the potential for surgical treatment. In clinical practice, patients with a large compressive goiter and a narrowed trachea are very common. The doctor and the patients are afraid of complications from surgery in this kind of goiter because the sternotomy is anticipated. This case reveals that trans-cervical thyroidectomy remains feasible.

ISSN No:-2456-2165

IV. CONCLUSION

Substernal goiters often present with compressive symptoms.

Chylothorax secondary to compressive goiter is an exceptional complication, Remainder surgery is the best treatment, when symptoms are present, removal of compressive goiter by a trans-cervical approach with thoracic duct repair may be an appropriate treatment to relieve symptoms without the need for a sternotomy.

Declaration of interests

The authors declare no conflict of interest

LISTS OF FIGURES

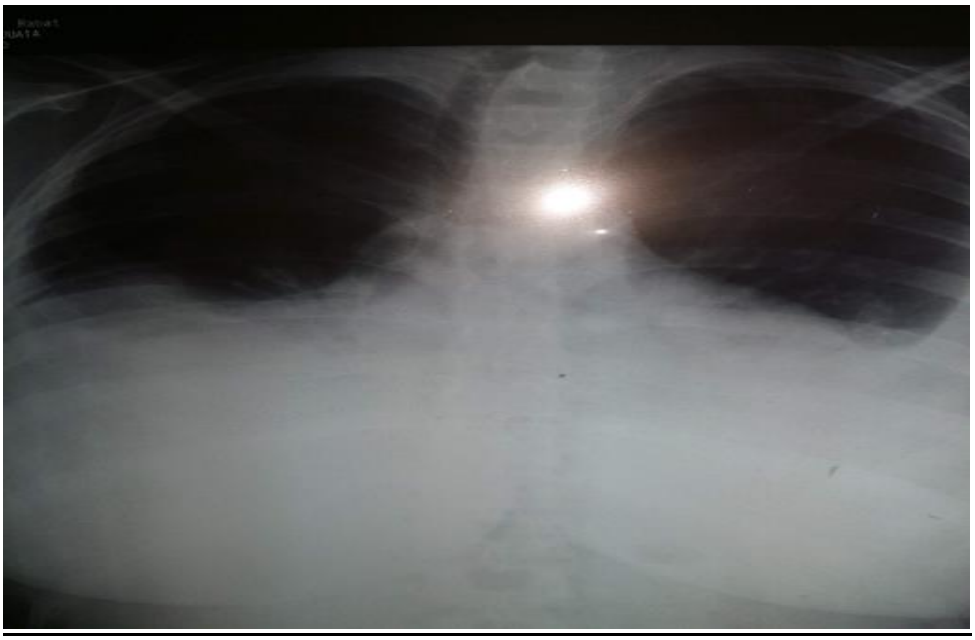

Figure 1: chest x-ray showing bilateral pleurisy More marked on the right

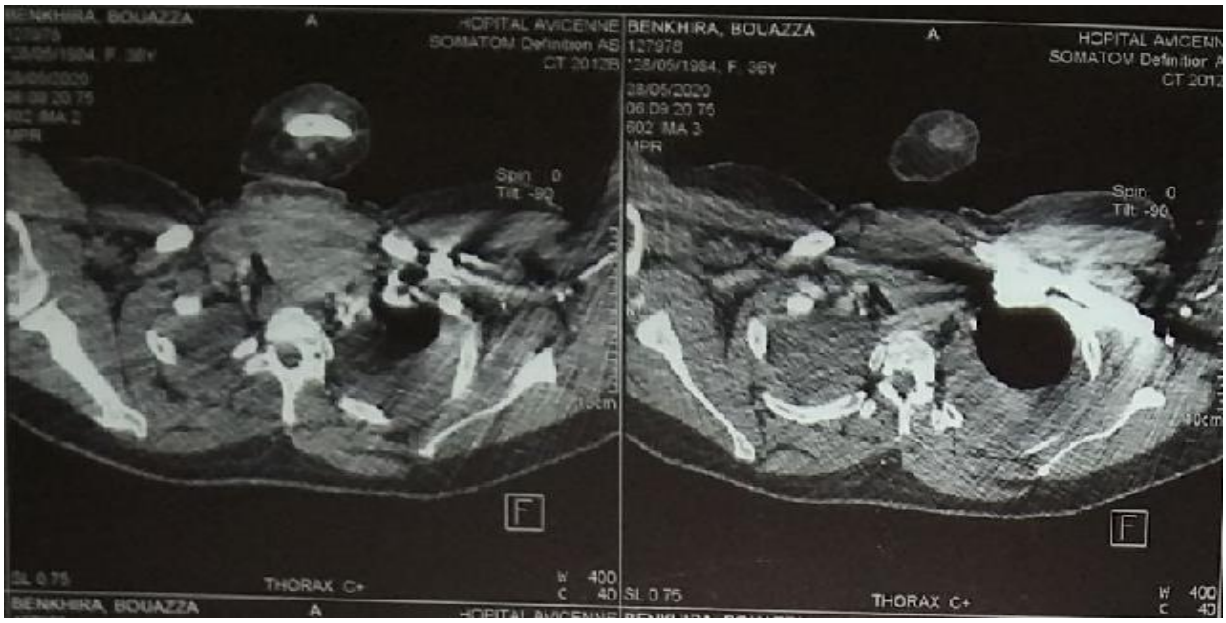

Figure 2: Cervico-thoracic CT showing a compressive goiter at the expense of the left lobe, pushing back the trachea and vascular structures

ISSN No:-2456-2165

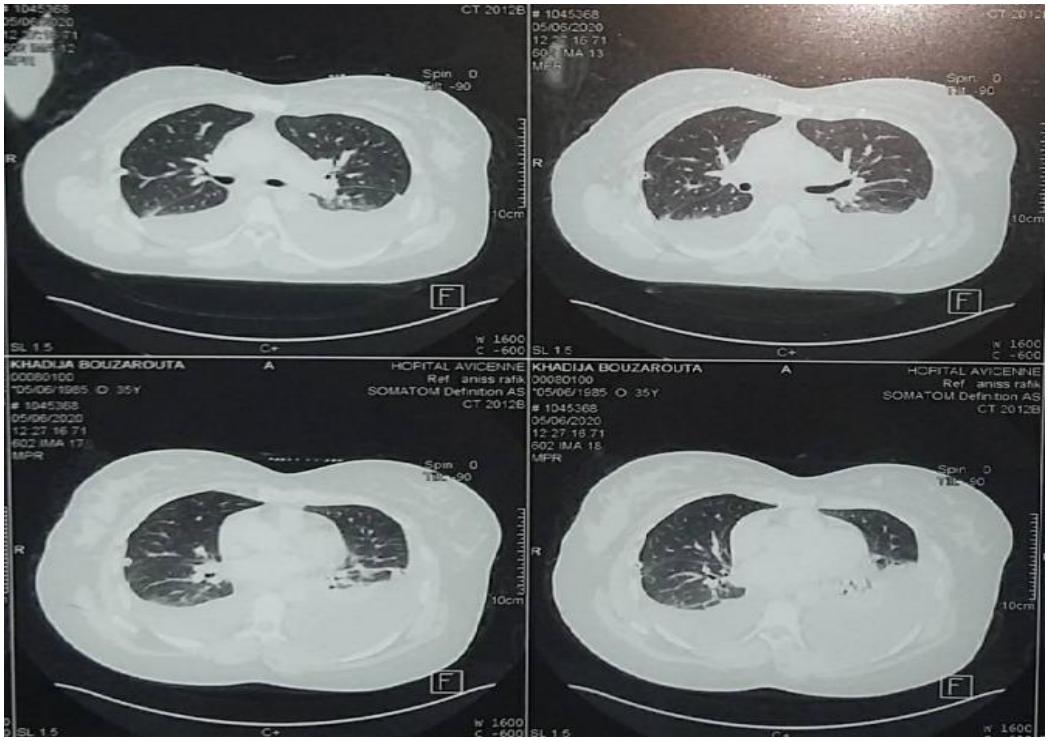

Figure 3: Chest CT showing bilateral pleural effusion

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