



Case Report / Olgu Sunumu

Parathyroid cyst: A rare benign mediastinal cyst

Paratiroid kisti: Nadir benign mediastinal kist

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ABSTRACT

Parathyroid cysts are extremely rare cystic formations in the mediastinum. Since the first described case, there have been approximately 150 cases reported in the English literature. Patients often present with symptoms related to compression of the adjacent tissues, while only 20 to 40% of cases have an evidence of hyperparathyroidism. For preliminary diagnosis, parathyroid scintigraphy can be helpful, although most cases are diagnosed by postoperative pathological analysis of surgical samples. The most optimal surgical procedure is total excision of the mass, where the surgical approach of choice can be either thoracotomy or median sternotomy according to the size and localization of the lesion. Herein, we report a 67-year-old male case of a mediastinal mass extending to the right hemithorax.

Keywords: Cyst; mediastinal; parathyroid.

Parathyroid cysts are rare lesions of the cervical region and the mediastinum. They were first described by Sandström, a Swedish anatomist.^[1] Mediastinal parathyroid cysts were first defined by Quervain in 1925.^[2] There have been approximately 150 cases of parathyroid cysts reported worldwide.^[3]

To date, five theories relating to the origin of parathyroid cysts have been proposed. These theories have suggested that parathyroid cysts originate from (i) embryologic remnants of the third and fourth branchial cleft; (ii) previously existing microcysts coalescing into one large cyst; (iii) simple retention of parathyroid secretions; (iv) atrophied remnants of the Kürsteiner canals of parathyroid glands of embryonic life; and (v) cystic degeneration of preexisting adenomas.^[4] However, none of these theories can adequately explain the pathophysiology of parathyroid

ÖZ

Paratiroid kistler, mediastende son derece nadir kistik oluşumlardır. İlk tanımlandığından bu yana, İngilizce literatürde yaklaşık 150 olgu bildirilmiştir. Hastalar genellikle kitlenin basısına bağlı semptomlar ile başvurmakla birlikte, hiperparatiroidi bulguları olguların yalnızca %20 ila %40'ında mevcuttur. Hastalığın ön tanısında paratiroid sintigrafisi faydalı olabilmekle birlikte, tanı genellikle cerrahi numunelerin ameliyat sonrası patolojik incelemesi ile konur. En uygun cerrahi işlem, kitlenin total rezeksiyonu olup, cerrahi seçenek lezyonun boyutuna ve yerine göre torakotomi veya median sternotomi olabilir. Bu yazıda, mediastinal kitlenin sağ hemitoraksa uzandığı 67 yaşında bir erkek olgu sunuldu.

Anahtar sözcükler: Kist; mediastinal; paratiroid.

cysts on its own, and it is likely that more than one mechanism is responsible for their formation.

Parathyroid cysts can be categorized into two as functioning and non-functioning.^[4] About 20 to 40% of cases present with signs of hyperparathyroidism and are accompanied by hypercalcemia symptoms such as nausea, vomiting, constipation, bone and joint pain, and pathological fractures.^[5,6]

In non-functioning cysts, due to the pressure on intra-thoracic organs, signs and symptoms such as dyspnea, dysphagia, vocal cord paralysis, or findings of thrombi caused by brachiocephalic vein or jugular vein compression are possible.^[6,7] Dysphonia is frequently seen in large mediastinal masses due to recurrent laryngeal nerve injuries.^[6] However, most mediastinal parathyroid cysts are asymptomatic.^[7]

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Figure 1. Thoracic computed tomography showing that a cystic mediastinal lesion extending to the compression of the trachea and esophagus.

Despite modern imaging techniques, mediastinal parathyroid cysts can be rarely diagnosed preoperatively. These cysts can be confused with bronchogenic cysts, duplication cysts, and thymic cysts.^[4] Parathyroid scintigraphy is valuable in determining the cysts originating from parathyroid gland. Parathyroid hormone (PTH) levels of the fluid inside the cyst being higher than serum levels are also useful for the diagnosis.

Herein, we report a 67-year-old male case of a mediastinal mass extending to the right hemithorax in whom the histopathological result was reported as a parathyroid cyst.

CASE REPORT

A 67-year-old male patient presenting with dysphagia complaints was referred to our Thoracic Surgery outpatient clinic. Previously, thoracic computed tomography (CT) revealed a mediastinal mass extending to the right hemithorax (Figure 1). The patient did not have symptoms such as myalgia, malaise, weight loss, polydipsia, or polyuria suggesting a preliminary diagnosis of hyperparathyroidism. He had a 30-pack-year history of cigarette smoking and was under follow-up for chronic obstructive pulmonary disease (COPD). There were no pathological findings on physical examination. In addition, preoperative laboratory test results were normal. The patient was scheduled for the excision of the cyst.

A written informed consent was obtained from the patient. The cyst covered with parietal pleura and surrounded by azygos vein inferiorly, trachea medially, superior vena cava anteriorly, and esophagus posteriorly was totally resected (Figure 2). The superior part of the cyst was connected to the thyroid gland; therefore, it was thought to be a parathyroid cyst. The PTH levels of the cyst fluid were not studied, since the test results would not alter the surgical procedure. Serum calcium and PTH levels were studied twice on Days 1 and 3, postoperatively. Calcium levels were 12.08 and 9.05 (reference range: 9-11 mg/dL), PTH levels were 32.56 and 34.84 pg/mL (reference range: 10-65 pg/mL), respectively, and all were reported to be normal. The patient was discharged on postoperative Day 5. He had no active complaints during follow-up in the postoperative first

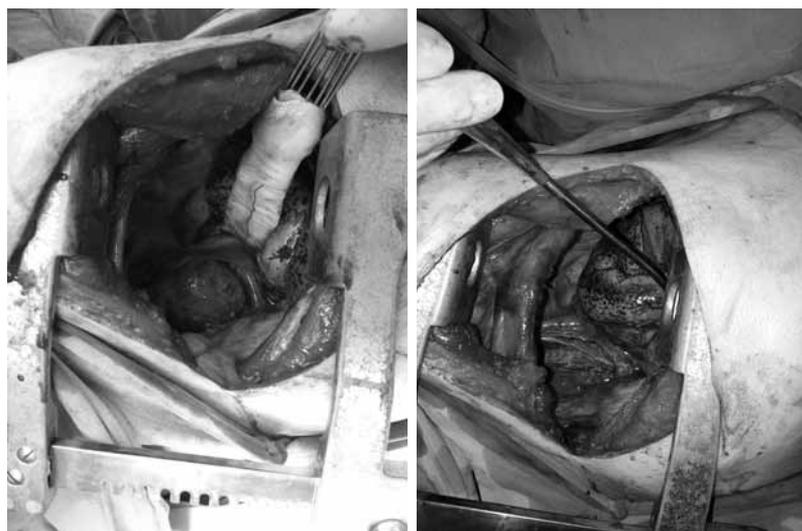


Figure 2. An intraoperative view of the cystic lesion.

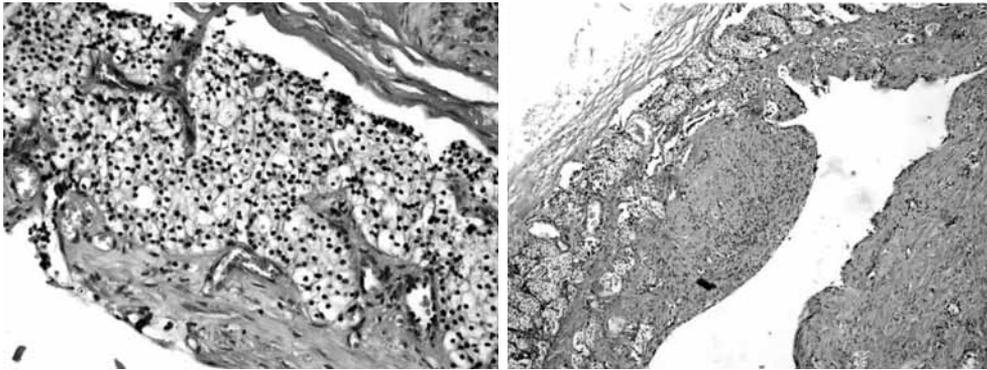


Figure 3. Image of hematoxylin and eosin staining of parathyroid cells in the cyst wall at $\times 5$ and $\times 20$ magnification.

and third months. Chest X-ray and blood work-up did not show any abnormal findings.

Gross examination of the surgical specimen showed a $7.5 \times 4.5 \times 1.8$ cm cyst and when it was cut open, the internal surface was septated. Immunohistochemistry profile was positive for pan-cytokeratin and cytokeratin 19, negative for vimentin, chromogranin A, and thyroid transcription factor 1, while the Ki-67 score was 1 to 3%. The histopathological result was reported as a parathyroid cyst (Figure 3).

DISCUSSION

Since they were first described in 1880, parathyroid cysts have been rare lesions which are mostly seen in the cervical region.^[2]

Functioning mediastinal parathyroid cysts of the mediastinum are easy to diagnose due to the presence of symptoms of hyperparathyroidism and an evidence of mediastinal cysts. However, non-functioning mediastinal parathyroid cysts, which are seen in up to 60 to 80% of cases, are usually diagnosed incidentally and most commonly following the surgical excision.^[5]

It should be kept in mind that mediastinal cysts can be parathyroid cysts. Pre- and intraoperative analyses of PTH levels of the intra-cystic fluid being higher than serum PTH level are valuable to confirm the diagnosis of parathyroid cysts. Yet, sampling of the cyst fluid before surgery may not be possible, due to location of the cyst, particularly in the mediastinum. In our case, PTH levels of the cyst fluid were not studied due to possible complications of percutaneous sampling of the cyst, and test results would not change the surgical procedure. Parathyroid cysts located in the mediastinum are most frequently in the anterosuperior mediastinal cavity (i.e., retrosternally above the innominate vessels) (60%), or in the retrotracheal space

(27%), and prevascular space (13%).^[6] In our case, the cyst was located in the anterosuperior mediastinal space.

As about 20% of mediastinal parathyroid cysts are accessible, they can be completely excised with a neck incision.^[7] For the rest, the choice of surgical approach can be modified according to the location and size of the mass. In our case, the patient could not tolerate one-lung ventilation with a double-lumen endotracheal tube due to preexisting COPD. In addition, video-assisted thoracic surgery was not possible; therefore, the surgical approach was a standard posterolateral thoracotomy. In recent years, video-assisted thoracoscopic surgery, robotic surgery, and the use of intraoperative gamma probe scanning have come into question.^[2] Furthermore, lately, it has been reported that robotic surgery is an effective and safe procedure for benign mediastinal lesions.^[8]

Total excision of the cyst is considered a curative treatment. Symptoms related to hyperparathyroidism and compression of the adjacent tissues completely resolve after the excision of the cyst.

The absence of specific findings on CT, magnetic resonance imaging, or ultrasonography makes it difficult to distinguish mediastinal parathyroid cysts from other mediastinal cystic lesions. As in our case, definitive diagnosis is usually made after the resection of the cyst.

In conclusion, parathyroid cysts should be considered in the differential diagnosis of mediastinal cystic lesions. If parathyroid cyst is suspected, the best diagnostic method is to look for parathyroid hormone level in cyst fluid and total excision of the cyst is considered a curative treatment.

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