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Giant cervicomediastinal non-functioning parathyroid cyst: A rare case

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Abstract
Parathyroid cysts of cervicomediastinal location are rare with only a few cases reported in the international literature. We present a rare case of parathyroid cyst in a 57 years old female with gradually increasing neck swelling and dysphagia. Examination revealed about 5 cm anterior neck swelling which moves with swallowing. Blood results including tumor markers and calcium levels were all unremarkable. Chest radiography noted visible mass with cervicothoracic sign and right tracheal deviation. A computed tomography showed a large anterior mediastinal cystic mass with left supraclavicular extension, size 5 x10 x 14cm pushing trachea to the right side from thyroid cartilage till carina level. She successfully underwent left subtotal thyroidectomy (for coexisting thyroid nodules) and median sternotomy and cyst excision. Operative histopathological examination revealed a parathyroid cyst. The patient's symptoms were relieved after surgery. Parathyroid cyst can therefore present as a rare cause of neck and mediastinal cystic lesion.

Keywords: Cervicomediastinal, non-functioning, parathyroid cyst, neck swelling

Introduction
Parathyroid cysts of cervicomediastinal location are rare with only a few cases reported in the international literature. We present a rare case of parathyroid cyst in a 57 years old female with gradually increasing neck swelling and dysphagia.

Case Report
A 57 years old female with hypertension and trigeminal neuralgia presented with gradually increasing anterior neck swelling for 10 years duration and mild dysphagia. Otherwise, no symptoms of hyper or hypothyroidism, odynophagia or dyspnea. No history of previous malignancy or any family members with malignancy. She had history of thyroid cyst excision more than 20 years ago. Physical examination noted mobile anterior neck swelling about 5 cm with no cervical lymphadenopathy. Previous neck incisional scar was well healed.

Blood investigations revealed her thyroid function test, AFP, CEA, CA 19.9, CA 125 and CA153 were all normal. Frontal chest radiograph showed a well-defined homogeneous opacity in anterior mediastinum with cervicothoracic sign that projected above both clavicles (Fig 1). Contrasted computed tomography of neck and thorax confirmed a large well defined anterior mediastinal mass with cystic component (CT attenuation of 10 Hounsfield unit, Fig 2a and 2b) extending from left thyroid and left supraclavicular region down to lower paratracheal region. Thyroid gland was displaced anterosuperiorly with clear demarcation but with multiple hypodense nodules within. The mass pushed the trachea to the right but no luminal narrowing of the airway. Clear demarcation with adjacent brachiocephalic vein, distal superior vena cava, left common carotid and aortic arch (Fig 2c).

Patient was informed that she had previous history of anterior mediastinal cystic mass aspiration about 2 years ago at a private centre and was told it was benign cystic content. However, no formal cytology examination available. Due to the presence of mild dysphagia and progressive enlargement of the swelling, the patient opted for definitive treatment with surgical resection.

After induction of general anesthesia, the patient was placed in supine position with neck extended. A 4 cm transverse cervical incision (about 2cm above sternal notch) was performed. Subsequently, anterior cervical strap muscles were divided and a thin-wall cyst was seen. Left subtotal thyroidectomy was performed by our otolaryngology colleagues in

order to aid for surgical mobilization of the cyst. We attempted to resect the cyst via cervical incision but failed in view of the cyst was deeply seated in middle mediastinum. Hence, we proceeded with median sternotomy and completed cystic mass dissection whilst keeping the entire mass intact (Fig 3). The cystic mass was separated from the surrounding structures (Anteriorly – Supra-aortic vessels and arch of aorta; Posteriorly – Tracheal and carina) and removed completely. Recurrent laryngeal nerves were preserved. A redivac drain was placed in cervical and mediastinal spaces followed by sternal and cervical wound closure.

The thyroidectomy specimen revealed two nodules one measuring 25mm diameter and another 3mm diameter, both of nodular hyperplasia on histological examination. The cystic mass measuring 110 x 85 x 15 mm, smooth and shiny, transparent, thin cyst wall less than 2mm thick with clear content (Fig 4). Microscopically, the cyst wall was lined by a single layer of cuboidal cells exhibiting bland round hyperchromatic nuclei with scanty cytoplasm while the stroma showed lobules of parathyroid tissue consisting of clear cells (Fig 6). No evidence of solid component, nuclear atypia or malignancy was seen. This was consistent with a parathyroid cyst.

The patient recovered well post-operatively but with improving hoarseness of voice due to left vocal cord palsy.

Discussion

Parathyroid cysts are rarely encountered neck masses. They are rare benign cystic masses of parathyroid gland occurring in about 1 to 5% of neck masses and 0.5 to 1% of parathyroid gland pathologies [1, 2]. Parathyroid cysts can be found anywhere from mandible down to mediastinum, but mostly found at the left thyroid lobe in 30% then superior mediastinum in 20% of the cases. Cervicomediastinal or cervicothoracic location occurred in less than 5% of the cases [2]. Inferior parathyroid glands are mostly affected and more localized on left side as in our case [2, 3].

In the literature, just over 350 cases of parathyroid cysts have been reported since the discovery of parathyroid glands by Ivar Sandström in 1880 [1, 2]. Most of the cases were reported from European countries [2, 4, 5], United States [6] or Latin America [3], Japan [7] or others but none particularly from South East Asian countries.

Several pathogeneses of parathyroid cysts have been postulated in the literature. They include coalescences of multiple microcysts, trapped secretion forming retention cysts, persistence of Kursteiner canals or embryological remnants of third or fourth branchial pouch [8]. In parathyroid adenomas, hemorrhage or infarction of the gland can lead to cystic degeneration of parathyroids [7]. Their sizes vary considerably from 0.5 to 15 cm, usually unilocular and solitary [1, 2].

Parathyroid cysts have a variable presentation [2]. Although parathyroid cysts are usually asymptomatic, they can cause hypercalcemia [2] or produce local compressive symptoms such as dysphagia [9], dyspnea or hoarseness of voice. It is more common in females in as much as 65% of the cases, peak incidence in the fourth and fifth decades of life [2].

Chest radiograph can show mediastinal widening and of interest, can suggest the origin of the mass based on the cervicothoracic sign [10]. A mass projecting above clavicle is less likely, and may extend to the neck if the cephalic border is obscured, as in our case. Ultrasonography of the neck can be helpful to check the mass characteristics and computed tomography will reveal a homogenous hypodense cystic mass and its relations to nearby structures.

As this case involved cervicomediastinal lesion, differentials include thyroid cysts, branchial cysts, thymic cysts, lipoma, teratoma or neurogenic and vascular tumors are all entertained. Due to its similar clinical characteristics and imaging features, clear distinction between thyroid and parathyroid cysts can be difficult [8]. Fine needle aspiration (FNA) could help ascertain the preoperative diagnosis of the parathyroid cyst with high parathyroid hormone levels detection in the clear watery aspirate [8], as compared to thyroid cyst fluids that would either be cloudy, gelatinous or bloody and negative for parathyroid hormone.

Treatment options include simple aspiration, sclerosant injection and surgical resection. In a functioning cyst or case of recurrence or symptomatic patients, surgery provides the best intent to cure. Careful complete excision of the mass without rupture is essential to prevent risk of recurrence, due to its very thin wall [2]. However, in cases of non-functioning cysts, simple aspiration can be attempted which can be both therapeutic as well as diagnostic, especially in the first presentation. Sclerosant therapy with intracystic injection of tetracycline or ethanol is another alternative but was hindered by the risks of neck pain, neurotoxicity and recurrent nerve palsy [5].

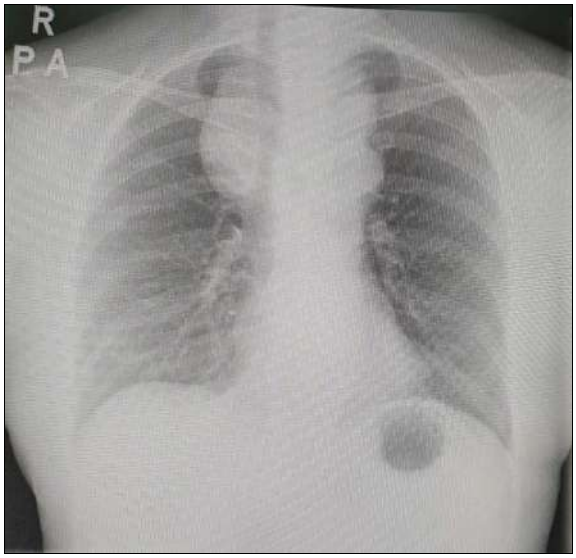

Fig 1: Chest radiograph: A well-defined homogeneous opacity in anterior mediastinum with cervicothoracic sign that projected above both clavicles. Trachea is deviated to right

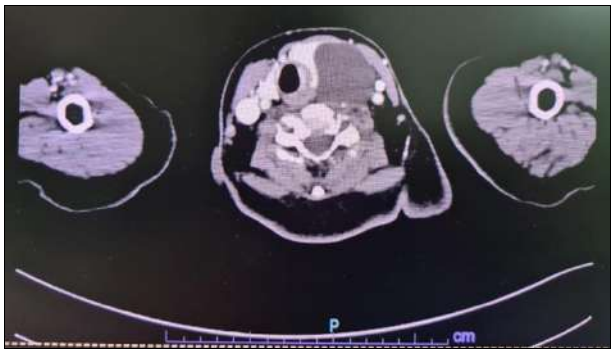

Fig 2a: Axial contrasted CT neck: large anterior mediastinal mass extends from left thyroid down to lower paratracheal region, displacing left thyroid lobe

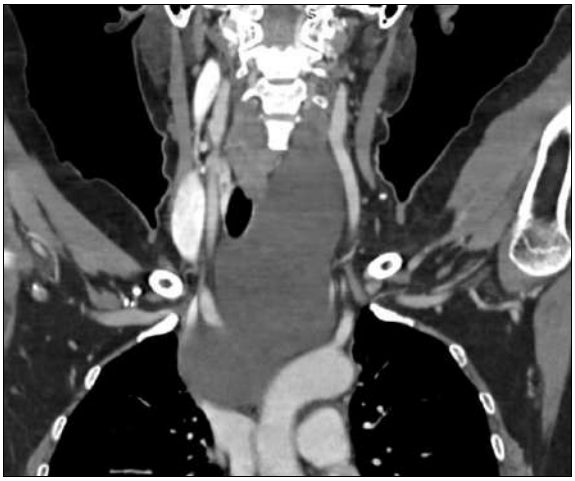

Fig 2b: Coronal contrasted CT neck: anterior mediastinal cystic mass has clear demarcation with adjacent brachiocephalic vein, distal superior vena cava, left common carotid and aortic arch

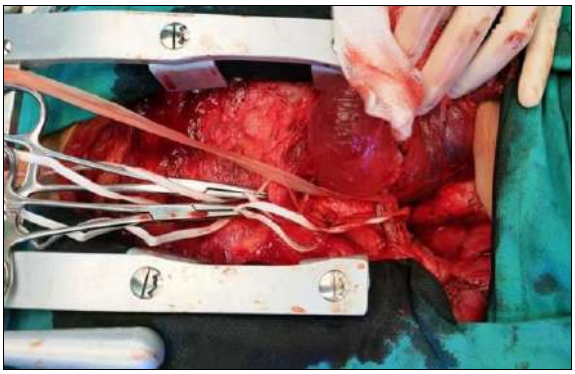

Fig 3: The cystic mass was separated from the surrounding structures (Anteriorly – Supra-aortic vessels and arch of aorta; Posteriorly – Tracheal and carina) and removed completely. Recurrent laryngeal nerves were preserved

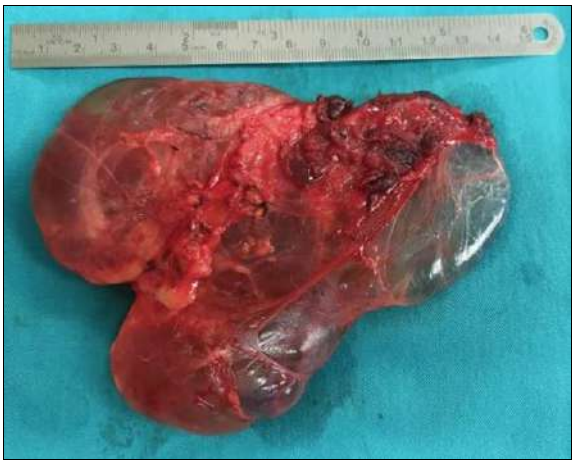

Fig 4: Parathyroid cyst excised intact, measuring 11 cm x 8.5cm, with clear colourless fluid content and shiny thin wall

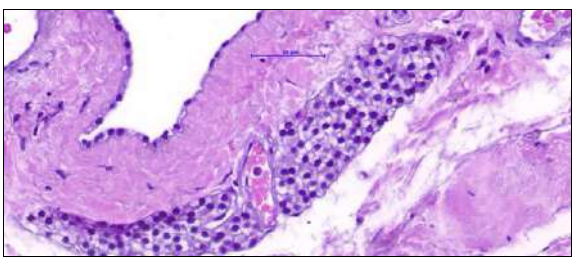

Fig 5: 400x magnification

Conclusion

To conclude, we described a case of a very large cervicomediastinal non-functioning parathyroid cyst which is an important but unusual cause of neck and mediastinal cystic lesions. Clinicians managing such cases must be aware of this possible diagnosis as complete surgical excision is the current best treatment. To our knowledge, this is the first case of parathyroid cyst in the South East Asia region.

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