

A Parathyroid Cyst; Often Neglected In The Differential Diagnosis Of Cystic Neck Swellings: A Case Report

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Abstract

Parathyroid cysts are a rare cause of cystic swellings of the neck. They are usually non-functioning and predominate in middle age women. We report the case of an apparently healthy 17-year-old girl with a relatively large cystic left sided neck swelling of two years duration. Diagnosis of Parathyroid Cyst was obtained after surgical resection. The case history, intraoperative finding and pathology were presented, the pitfalls of diagnosis were highlighted and the condition was discussed with appropriate conclusions and recommendations.

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INTRODUCTION

Parathyroid cysts (PCs) are rare lesions, predominant in females and usually located in the neck but may be located in the mediastinum.⁽¹⁾

They are frequently a pitfall diagnosis of thyroid pathologies; usually asymptomatic and frequently omitted in the differential diagnosis of neck masses.⁽²⁾ Its importance lies in the diagnostic difficulty and the final diagnosis is usually made during surgery confirmed by histopathological examination.⁽³⁾ A crystal-clear fluid and an elevated Parathormon Hormon (PTH) level in cyst fluid confirm the diagnosis.⁽³⁾

The majority of the lesions are non-functioning; however, functioning cysts are associated with

hyperparathyroidism.⁽⁴⁾ It is also possible to cause compressive symptoms and even recurrent laryngeal nerve palsy.⁽⁴⁾

Till 2012; there has been around 300 cases described in the literature. The exact incidence is still discussed, given that in a recent study, review of its occurrence ranged from 0.08 to 3.4% of the cases of thyroid or parathyroid resections.⁽⁵⁾

In the present paper; we are reporting a case of PC in a 16-year-old girl. The case history, intraoperative finding and pathology shall be presented, the pitfalls of diagnosis shall be highlighted and the condition shall be discussed.

CASE HISTORY

An apparently healthy 17-year-old girl was seen in Dec. 2014 with a history of progressive painless enlargement of a swelling over the last two years which was occupying the lower left side of the neck. Recently; she started uncomfortable feeling in her neck but no specific complaint concerning breathing or swallowing. She had been seen few months ago by a surgeon who aspirated the swelling twice and the mother admitted that it soon recollect every time. On examination; the swelling is cystic, non-tender, hardly transilluminable and slightly moves with swallowing. There was no clear clinical evidence of retrosternal extension. No clinical evidence of palpable cervical lymphadenopathy and the rest of the general examination was unremarkable (Figure 1).



Figure 1: A cystic swelling of the neck in a 16-year-old girl

The mother refused a trial of aspiration and requested surgery. Ultrasound (U/S) of the neck reported a cystic swelling intimately related to the left thyroid lobe which was announced as "normal" so is the right thyroid lobe and the isthmus along with negative lymph node enlargement. The Chest X-ray and the Magnetic Resonance Imaging (MRI) of the neck are presented in Figure 2(A and B) respectively. MRI reported the cystic swelling within the left lobe of the thyroid gland with some retrosternal extension posterior to the medial ends of the clavicles. Tracheal deviation to the opposite side was also documented.



Figure 2: (A) chest x-ray; the trachea is seen pushed to the right side.



Figure 2: (B) MRI scanning of the neck showing clear delineation of the cystic swelling

Intraoperatively; a large cystic lesion measuring 10x6 cm was detected. The wall is thin and the content is aqueous. It was seen exactly at the site of left thyroid lobe completely obscuring the field. After full mobilization of the cyst, a cleavage plan could be seen between the cyst and the small normal looking left thyroid lobe. No trial to dissect the left thyroid lobe was performed. The cyst ruptured on attempting removal intact and the fluid spilled was water-clear. The intraoperative findings are demonstrated in Figure 3 (A and B).

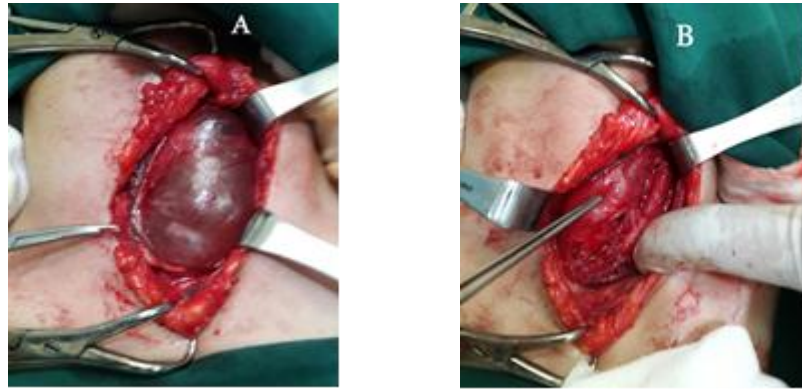


Figure 3: (A) the view of the cyst through a lower transverse cervical incision. (B) After removal of the cyst; the forceps is pointing to the normal-looking left thyroid lobe

The excised cyst was submitted to histopathological assessment and its relation to parathyroid tissue was documented. Microscopically; the cyst wall is composed of fibrous tissue with entrapped nest of parathyroid cells (Figure 4 A and B).

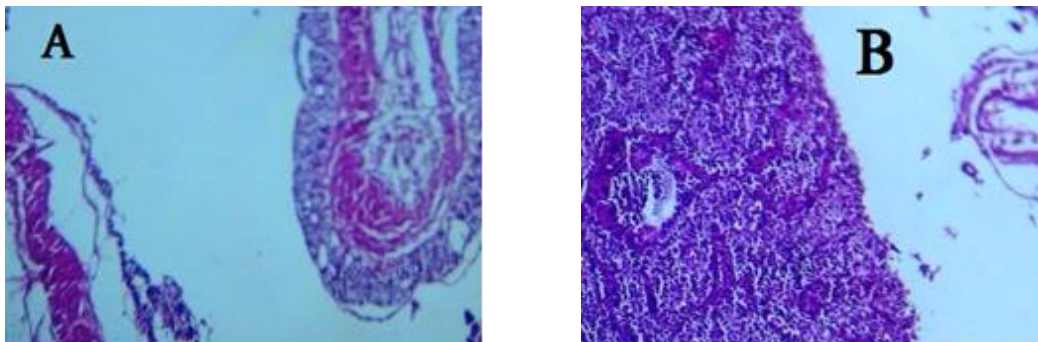


Figure 4: (A) cyst wall lined by parathyroid cells. (B) Showing parathyroid tissue adjacent to cyst wall

The patient had an uneventful recovery. Postoperative PTH and serum calcium levels were normal and documented for follow up. The patient was seen 6 months later and clinical examination, U/S and biochemical assessments were all normal..

DISCUSSION

In 1880; Sandstorm; a Swedish anatomist, first described a macroscopic PC and in 1905; Goris; a Belgian surgeon, first described a surgical removal of PC.⁽⁶⁾ The first case of a PC that had been diagnosed by Fine Needle Aspiration Cytology (FNAC) was reported by Crile Jr. and Perryman in 1953.⁽⁷⁾ The rarity in the number of documented cases is attributed to the lack of recognition of these cysts in clinical practice.⁽⁸⁾

Referring to surgical studies; in 2009; McCoy et.al reported a 3% incidence of cystic parathyroid lesions in 1769 patients undergoing parathyroidectomy for primary hyperparathyroidism, however; in a recent retrospective analysis of 6621 patients submitted to neck U/S, Cappelli et.al reported a 0.075% incidence in an unselected population which was much lower than previously reported.^(9,10)

PCs occur in both genders, with a male: female ratio of 1:2.5 normally at the 4th and 5th decades of life, however; to date, several pediatric cases have also been described.

The vast majority is located in the lower parathyroid glands, described from the angle of mandible to the mediastinum with slight preponderance on the left side. Most pure PCs are orthotopic and solitary and are found more frequently in women. Nevertheless; it can be ectopic and localized anywhere in the cervical region, including the thyroid, the mediastinum and the thymus. 10% of cases occur in the mediastinum, usually the anterior region.⁽⁸⁻¹⁰⁾

Pathogenesis of a parathyroid cyst is uncertain, and there are some theories that explain its origin; it can be an embryological remnant of the 3rd and 4th branchial arches causing increase in volume by accumulation of secretions and forming a macrocyst. It can also result from degeneration, infarction or haemorrhage in a parathyroid or in an adenoma of a parathyroid gland^(11, 12). It can be the result of Coalescence of microcysts of a normal or adenomatous parathyroid. It may be the result of Persistence of "Kustneiner canal" which is a remaining of fetal life. Retention of PTH in colloid vesicles was also implicated as a possible factor.^(11,12)

PCs are of two types; non-functioning cysts are described as the most common (around 90%) and predominant in females with a mean age of 43.3 years. It usually affect inferior parathyroid and presents as an asymptomatic neck mass.⁽¹³⁾ PTH in cyst fluid is raised. Histopathologically; they are simple cysts lined by a flattened cubic to low-columnar epithelium. Several types of parathyroid cells are found in their walls like chief cells, water-clear cells and oxyphil cells.⁽¹³⁾

Functioning PCs comprise the rest of 10% and occur in variable locations. They are usually associated with hyperparathyroidism. Rarely; it can be the cause of malignant Hypercalcaemia with primary hyperparathyroidism⁽¹⁴⁾. Patients are usually male (1.6 times more frequent than in females) and of older age (a mean of 51.9 years). PTH in cyst fluid is markedly raised.⁽¹⁵⁾ Histopathologically; some functional cysts lack an identifiable lining and are more properly termed pseudo-cysts. They may contain foci of haemorrhage or necrosis. It is believed that these may arise from degenerating adenomas. Other functional cysts are multilocular, complex lesions with thin walls and are found in close association with either an adenoma or a hyperplastic gland.⁽¹⁵⁾

The heterogeneous clinical presentation of PCs is determined by their hormone activity, size and location. The differential diagnosis includes thyroglossal duct cyst, branchial arch cyst, thyroid adenoma and Parathyroid carcinoma. Non-functioning cysts are usually discovered incidentally during

physical examination, imaging evaluation for other reasons and after surgical excision of thyroid nodules and histological examination.⁽¹⁵⁾ Large non-functioning cysts may be localized in the mediastinum and it can mimic retrosternal goitre causing compressive symptoms such as dysphagia, dyspnoea, vocal cord palsy, respiratory failure and jugular vein thrombosis.⁽¹⁶⁻²⁰⁾ Recently; a giant cystic adenoma masquerading as a retropharyngeal abscess, had also been reported.⁽²¹⁾ It can also be located inside the thyroid where they are discovered incidentally on FNA during evaluation of a multinodular goiter or as a solitary thyroid nodule.⁽²²⁾

FNA remains a valuable diagnostic tool. Typical findings of watery colorless crystal-clear fluid are suggestive of the diagnosis in the majority of non-functioning cysts, although a colored aspirate does not exclude the existence of a parathyroid cyst.⁽²³⁾ In some cases of functioning parathyroid cyst, the aspirate may be yellow, brown and even blood-stained due to pre-existing degenerated or infarcted parathyroid adenoma containing Haemosidrin. The diagnosis can be established by elevated level of PTH in the aspirated fluid.⁽²³⁾ A thyroid cyst yields a yellowish or bloody aspirate with high level of thyroglobulin and undetected level of PTH while a branchial cyst yield yellowish viscous aspirate with no detectable hormones. In this context, cytology of aspirated fluid is not particularly helpful.⁽²⁴⁾

U/S describes a cystic anechoic structure with thin wall and posterior hypoechoic enhancement. CT scan and MRI; particularly helpful in delineating the anatomy in large cysts, demonstrate only neck mass with cystic contents and its anatomical relations. Thyroid scan may reveal the presence of a non-functioning cyst but it does not distinguish if the lesion is in the thyroid or in the parathyroid gland.⁽²⁵⁾

Concerning general outlines of management; nonfunctioning cysts without complications can be aspirated under U/S guidance. The early complication of post-evacuation is haemorrhage inside the cyst, while late complication is the recurrence of the cyst. In both situations; percutaneous evacuation can be repeated. Recurrent cyst should be treated surgically.⁽²⁶⁾ When there is an intraoperative suspicion that the pathology is related to the parathyroid; a trial to inspect other parathyroid glands is recommended.^(12, 26)

Conclusions and recommendations:

Regarding such pathology; this case report exemplifies the preoperative clinical diagnostic difficulties thus confirming literature data and drawing attention to the importance of including this possibility in any differential diagnosis for a cystic neck swelling. Aspiration of the cyst fluid and assessment for PTH is diagnostic and highly recommended as far as a preoperative correct diagnosis is essential to the therapeutic decision.

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