Parathyroid adenoma in third pharyngeal pouch cyst as a rare case of primary hyperparathyroidism

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ABSTRACT

The primitive thymus and inferior parathyroid derive from the third branchial cleft. During embryonic development, these structures descend, reaching their final localisation. Third branchial cleft anomalies present usually as a fistula, abscess or cyst. However, there are no reports on parathyroid adenomas in the literature other than as a morphological possibility.

We describe the case of a 47-year-old man, who had been diagnosed with arterial hypertension and who presented with a cervical mass at the edge of the lower third of the sternocleidomastoid muscle. On ultrasonography, the mass had a cystic walled appearance. Laboratory analysis only revealed an intact parathyroid hormone level of 140.5pg/ml. Sestamibi imaging showed a probable parathyroid adenoma in the anterior mediastinum. During surgery, a tract running from beyond the superior thyroid pedicle to the superior mediastinum was dissected and removed. In the inferior end of the tract, a brown mass was visible. Pathological examination revealed a thymus cyst surrounding a parathyroid adenoma. The primal alteration was the lack of division between the thymus and inferior parathyroid gland, and the prompt prevention of their development. In the case of our patient, a parathyroid adenoma had grown by chance.

KEYWORDS

Hyperparathyroidism – Parathyroid glands – Parathyroidectomy

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Most of the cervical area structures derive from primitive pharyngeal (or brachial) apparatus, which is organised into pouches, arches and clefts. This pattern is a transient embryonic structure that will be obliterated completely by the seventh embryonic week. Among all of these embryonic structures is the third pharyngeal pouch, where the primitive thymus and inferior parathyroid originate.¹ During embryonic development, when the thymus migrates caudomedially into the superior mediastinum, it pulls the inferior parathyroid to rest on the dorsal surface of the thyroid gland. The third pharyngeal pouch abnormalities usually appear as a fistula, abscess or cyst but are quite uncommon (less than 5%).² However, no parathyroid adenomas have been reported in the literature other than as a morphological possibility. We present the case of a middle aged man with a neck mass and a parathyroid hormone (PTH) elevation.

Case History

A 47-year-old man diagnosed with arterial hypertension was referred to our department reporting a painful mass in the neck that had never been appreciated prior to the onset of pain. He denied a history of trauma or previous

infection. Physical examination revealed a cervical mass at the lower edge of the left sternocleidomastoid muscle. No evidence of cervical adenopathy was presented. The mass presented clinically as a smooth, tender, soft, solitary lump.

Laboratory analysis revealed a total calcium level of 2.47mmol/l, a parathyroid hormone (PTH) level of 140.5pg/ml, a phosphorous level of 0.807mmol/l, a thyroid stimulating hormone level of 1.65µiu/ml, a vitamin D level of 9.72nmol/l, negative antithyroid microsomal antibodies and negative antithyroglobulin antibodies. Ultrasonography demonstrated a cystic walled appearance. The mass was located laterally and immediately inferior to the left thyroid lobe, indicating an intrathoracic component (Fig 1). Magnetic resonance imaging showed the mass as rich in protein or haematic fluid, and its location was posterior to the left thyroid lobe and penetrated into the anterior mediastinum (Fig 2). Sestamibi imaging showed a probable parathyroid adenoma in the anterior mediastinum. Fine needle aspiration was performed and cyst fluid was sampled. The PTH assay of the fluid was negative and cytology only revealed macrophages.

A decision was made to treat the hyperparathyroidism surgically. The patient was taken to the operating room where a duct was discovered, running from the cystic

LSCM = left sternocleidomastoid; TPC = third pouch cyst

Figure 1 Ultrasonography showing a third pouch cystic walled mass in contact with the medial aspect of the left sternocleidomastoid muscle

Figure 2 Magnetic resonance imaging showing a mass (third pouch cyst [TPC]) posterior to the left thyroid lobe and penetrating into the anterior mediastinum

lesion to beyond the superior left thyroid lobe pole (Fig 3). A brown mass could be seen inside the cystic lesion (Fig 4). The duct was close to the carotid artery and internal jugular vein so careful dissection was performed (Fig 5) and the surgical specimen was obtained. Pathological diagnosis

Figure 3 Intraoperative photograph: A duct (D) was discovered running from the cystic lesion (CL) and towards the superior left thyroid lobe pole and beyond.

Figure 4 Intraoperative photograph: A brown mass suspicious of a parathyroid adenoma (PA) could be seen through the cyst wall.

confirmed a thymus cyst, which contained a parathyroid adenoma.

The two-year follow-up period has not revealed any recurrence of hyperparathyroidism. A laboratory test showed a total calcium level of 2.06mmol/l and a PTH level of 63.7pg/ml. Only hypertension remains despite having removed the parathyroid adenoma.

Discussion

In this case, the primal alteration was the lack of division between the thymus and inferior parathyroid gland, and the prompt prevention of their development. A fact that

Figure 5 Intraoperative photograph: The duct (D) was close to the carotid artery (CA) and internal jugular vein (IJV).

supports this theory is the close relationship found between the duct and the carotid artery in the operating room. Both elements derive from the primitive third arch and their development is closely associated. This alteration leads to the persistence of the thymopharyngeal duct, which runs lateral to the pharynx on the inferior constrictor muscle, deep to the vessels and nerves, and also deep to the infrahyoid muscles, behind the lateral lobe of the thyroid, entering the superoanterior mediastinum.² A parathyroid adenoma developed in the altered parathyroid gland of our patient, adding even more interest to this unusual pathology.

No other cases have been found in the literature concerning third pouch cysts with a parathyroid gland inside

with clinical similarities such as neck mass as the first symptom, location of the mass, absence of previous antecedents and a surgical common dissection. In the case described by Charous *et al* of a thymic cyst, the inferior parathyroid gland was normal.⁵ A combination of a thyroglossal duct cyst and a parathyroid gland has also been reported.⁴ This could mimic the clinical presentation of our case. Furthermore, some authors have gathered small series of parathyroid cysts, which could present as hyperparathyroidism with similar ultrasonography results. Some of the patients in the work by Armstrong *et al* presented with parathyroid cystic lesions in the thyrothymic ligament.⁵ This could mimic the abnormal location of the parathyroid lesion in our patient.

Conclusions

Although comparable clinical cases can be found, the case we report shows a third pouch persistence with a thymic cyst and a parathyroid gland associated in a patient with hyperparathyroidism.

References

1. Mirilas P. Lateral congenital anomalies of the pharyngeal apparatus: part I. Normal developmental anatomy (embryogenesis) for the surgeon. *Am Surg* 2011; **77**: 1,230–1,242.
2. Mirilas P. Lateral congenital anomalies of the pharyngeal apparatus: part II. Anatomy of the abnormal for the surgeon. *Am Surg* 2011; **77**: 1,243–1,256.
3. Charous DD, Charous MT, Cunnane MF, Spiegel JR. A third branchial pouch cyst presenting as a lateral neck mass in an adult. *Ear Nose Throat J* 2006; **85**: 754–757.
4. Cheng CY, Chang YL, Hsiao JK, Wang CP. Metachronous thyroglossal duct cyst and inferior parathyroid cyst: a case report. *Kaohsiung J Med Sci* 2008; **24**: 487–491.
5. Armstrong J, Leteurtre E, Proye C. Intraparathyroid cyst: a tumour of branchial origin and a possible pitfall for targeted parathyroid surgery. *ANZ J Surg* 2003; **73**: 1,048–1,051.