

Parathyroid cysts, a rare condition: a case report and review of the literature

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Rezumat

Chisturile paratiroidiene, o situație clinică rară: prezentare de caz și discuții

Chisturile paratiroidiene reprezintă o adevărată încercare clinică și terapeutică pentru medicul practician. De cele mai multe ori, dimensiunile acestora sunt mici, de câțiva milimetri în diametru, deși uneori pot atinge dimensiuni centimetrice. În funcție de activitatea secretorie, există 2 tipuri de chisturi: forme clinice nonfuncționale (esențiale) și forme funcționale (adenomatoase), ultimile fiind responsabile de hiperparatiroidism. Autorii prezintă în acest articol un caz clinic de chist paratiroidian nonfuncțional, simptomatic, la o pacientă de 36 de ani, prezentându-se clinic ca o tumefacție cervicală anterioară. Sunt expuse de asemenea metodele de diagnostic preoperator și conduita terapeutică actuală.

Cuvinte cheie: paratiroidă, chist, prezentare de caz

Abstract

Parathyroid cysts are a rare medical condition, that is why they represent a diagnostic and therapeutic challenge for the practitioner. The cysts are often small in diameter, measuring a few millimeters, but in some cases they are centimetric in size. There are 2 types of parathyroid cysts, according to their activity: the nonfunctioning (essential) and the functioning

(adenomatous) forms, the last one being responsible of hyperparathyroidism. We report a case of nonfunctioning symptomatic parathyroid cyst in a 36-year old woman revealed by an anterior cervical tumefaction focusing on the pre-operative diagnosis and the management of it regarding the various therapeutic methods described in the literature.

Key words: parathyroid, cyst, case report

Introduction

Parathyroid cysts are rare neck tumours. Though they were discovered in 1880 by Sandstrom and excised for the first time in 1906 by Goris, only about 250 cases were described in the literature by now. These cysts usually occur during the fourth and fifth decade of life and are defined in relation to their parathyroid hormone(PTH) secretion capacity: nonfunctioning (essential) and functioning (adenomatous) types. The non-functioning types (80% of cases) are more common in females, while the functioning types are more common in males. (1) A single case of malignant transformation has been described. (2)

The non-functioning parathyroid cysts' origin is still under debate and there are several hypotheses to explain this:

- forms resulting from the fusion of small cavities within the parathyroid parenchyma;
- vestigial cysts resulting from the 3rd and 4th branchial pouches;
- pseudocysts caused by the parathyroid's necrosis, containing high levels of PTH but not accompanied by an increase of plasma levels of the hormone.

The functioning parathyroid cysts' origin is generally

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accepted as the result of cystic degeneration of a parathyroid adenoma.

In most cases the parathyroid cysts are asymptomatic (3). Sometimes the cysts are revealed by a tracheal or recurrent nerve compression (4). The symptoms associated with the adenomatous cysts are related to excessive secretion of PTH.

Physical examination usually shows a soft, mobile swelling often located at the inferior pole of the thyroid. There are cases of multiple cysts described and even cysts developed at other cervical and mediastinal levels (5,6). Cervical ultrasonography (US), followed in some cases by computer tomography (CT) and magnetic nuclear resonance (MRI) shows a cystic, juxtathyroid swelling with paper-thin walls. The differential diagnosis includes a thyroid cyst, a parathyroid cyst and a branchial cyst. The diagnostic dilemma can be solved after the fine-needle aspiration of the cyst and examination of the intracystic fluid. In parathyroid cysts it is a watery, colourless, acellular liquid, with high levels of PTH for both nonfunctioning and functioning types (3,4,7). PTH in the cystic fluid is almost entirely in the form of the non-active C-terminal fragment, since PTH is rapidly broken down. In the adenomatous forms, there are the same high PTH levels in the cystic fluid but in addition the serum PTH (active) levels are high due to secretion by the adenomatous cells.

Case report

D.M., a 36-year old female with no particular antecedents, presented in our department in 2008 with a 3-year history of anterior cervical tumefaction, located at the inferior pole of the right thyroid lobe. The patient accused a discrete dysphagia and dispnoea upon effort. The tumefaction was roundish, soft, about 3,5 cm in diameter, not painful during palpation. Laryngoscopic findings were normal.

Cervical US showed a hypoechoic, oval-shaped formation with well defined margins and thin walls, in close contact with the posterior part of the right thyroid lobe (Fig. 1).

The thyroid gland hormones' levels were normal as well as the phosphocalcic metabolism markers. The serum PTH level was 28,8 pg/mL (normal values 11-47 pg/mL).

Fine needle-aspiration of the cervical tumefaction revealed 10 ml of clear, acellular liquid. The biochemical study of the liquid showed high levels of PTH (426 pg/mL).

3 weeks after the treatment by fine-needle aspiration, the patient returns with the same cervical tumefaction that seems to be greater in size than previously. We decide the surgical management of the tumefaction under general anesthesia. The cyst located at the level of the inferior right parathyroid was easily dissected without opening the capsule and then removed along with the right inferior parathyroid gland (Fig. 2).

The surgical specimen measured 57 x 30 x 24 mm. Histological examination showing a parathyroid cyst with a capsule adhering to the parathyroid parenchyma and fibroadipose tissue, along with previous laboratory tests concluded of a non-functioning parathyroid cyst.

Postoperative care was simple, the patient leaving the hospital the next day. The patient was examined 3 months

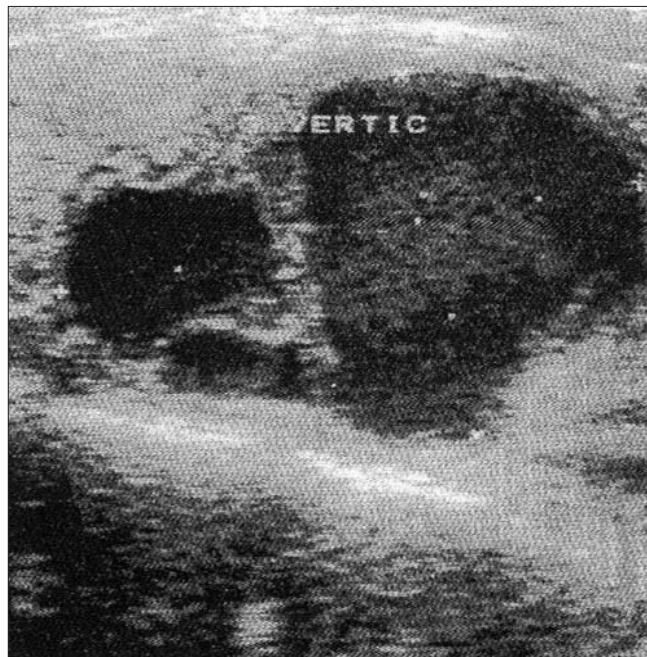


Figure 1. Cervical ultrasonography. Hypoechoic formation with well defined margins in close contact with the posterior part of the right thyroid lobe



Figure 2. View during surgical removal of the cyst. Parathyroid cyst located at the level of the inferior right parathyroid gland

later with no clinical or ultrasonographic evidence of relapse.

Discussion

Although parathyroid cysts were found in 42 cases from 100 autopsies by Black and Watts (8) suggesting a quite frequent pathology this is the only documented case of parathyroid cyst managed in our department in a 10-year period.

In 1979, Calandra and al. (9) found 11 (3,38%) functional parathyroid cysts among 325 patients hospitalized in the

surgery department for hyperparathyroidism. In another article, Clark (10) reviews this particular pathology and finds out in the literature that among 96 parathyroid cysts there were only 14 functional cysts (14,6%).

Our case highlights the management of a non-functional parathyroid cyst taking into consideration the more recent reports in the literature, beginning with a cervical ultrasonography and a fine-needle aspiration of the cystic fluid for the cytological assessment. In case of a clear, acellular, colorless fluid one must be aware of the possibility of a parathyroid cyst. Also, the high levels of PTH in the fluid are in correlation with this diagnosis whatever the type of the cyst. Moreover, laboratory tests reveal if the cyst is nonfunctioning (normal PTH, calcium and phosphorous serum levels - as in our case) or functioning (high serum levels of PTH, calcium and phosphorous).

Fine-needle aspiration leads frequently to a relapse in 2 cases out of 4 in one month time in Prinz et al.'s experience (11) and in 2 cases out of 12 in Shi's experience (12). Some authors (13) suggest a tetracycline injection in the cyst as an alternative to the surgical treatment, but this method has a high risk of cyst's wall rupture followed by intense cervical pain (14,15) or even recurrent laryngeal nerve palsy (16).

In case of a nonfunctioning cyst, Clark suggests the surgical management of the cyst after the first relapse of the ultrasonography-guided fine-needle aspiration, in case of patients complaining of significant symptoms or in case of other locations (mediastinal, retromandibular). In case of a functioning cyst the surgical excision of the cyst is always indicated as the first option.

New techniques such as minimally invasive endoscopic parathyroidectomy represent an alternative to the classic surgical approach and are yet to be evaluated.

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