



Surgical treatment of a functional parathyroid cyst in a patient with primary hyperparathyroidism: A case report

Elena A. Ilyicheva^{a,b,*}, Gleb A. Bersenev^a

^a Federal State Budgetary Scientific Institution "Irkutsk Scientific Center of Surgery and Traumatology", ul. Bortsov Revolyutsii 1, Irkutsk, Irkutsk Region, 664003, Russian Federation

^b State Budgetary Institution of Public Health "Irkutsk Regional Clinical Hospital", Mikrorayon Yubileyniy 100, Irkutsk, Irkutsk Region, 664049, Russian Federation

ARTICLE INFO

Article history:

Received 26 November 2020

Received in revised form

30 November 2020

Accepted 1 December 2020

Available online 4 December 2020

Keywords:

Parathyroid cyst

Primary hyperparathyroidism

Case report

Surgical treatment

ABSTRACT

INTRODUCTION: The frequency of parathyroid cysts in patients with primary hyperparathyroidism is from 1 to 2 %. Nowadays, parathyroid cysts remain a difficult diagnostic and therapeutic task.

PRESENTATION OF CASE: A 57-year-old woman was referred to an endocrine surgeon to evaluate a presence of a mass in the left lateral region of the neck, the appearance of shortness of breath. In the biochemical analysis of the patient's blood, the serum ionized calcium level was increased - 1.53 mmol/l, parathyroid hormone - 149.6 pg/ml. Ultrasound and MSCT scan of the neck showed the presence of a fluid formation measuring 80 × 40 × 33 mm, which was displacing the trachea medially to the right. Fine-needle aspiration revealed the level of iPTH in cystic fluid >2500 pg/mL. The patient underwent cervicotomy, left lower parathyroidectomy with parathyroid cystectomy. According to a histological study, the cyst wall was presented by fibrous tissue, which was lined with epithelium from the dark main cells of the parathyroid gland. Remission of primary hyperparathyroidism was achieved.

DISCUSSION: This clinical report confirms the literature on a decrease in the sensitivity of imaging methods in the diagnosis of parathyroid cysts. A decrease in the effectiveness of intraoperative monitoring of parathyroid hormone with parathyroid cysts was confirmed.

CONCLUSION: With parathyroid cysts, a comprehensive assessment of all imaging methods is required. Functional parathyroid cysts require surgical treatment. Intraoperative monitoring of iPTH has low sensitivity in relation to the detection and assessment of the radicality of the operation for parathyroid cysts.

© 2020 Published by Elsevier Ltd on behalf of IJS Publishing Group Ltd. This is an open access article under the CC BY-NC-ND license (<http://creativecommons.org/licenses/by-nc-nd/4.0/>).

1. Introduction

Parathyroid cysts are a rare endocrine pathology [1]. Their prevalence is 0.5 % among pathologies of the parathyroid glands and 1–5 % of all neck tumors [2]. This pathology was first described in 1880 by I. Sandström, a scientist who discovered the parathyroid glands [3]. D. Goris in 1905 published a report of the first successful case of surgical treatment of a parathyroid cyst [4]. Fewer than 400 cases of parathyroid cysts have been reported since the first description in the second half of the 19th century [5].

Depending on the ability to synthesize parathyroid hormone and cause hyperparathyroidism, parathyroid cysts are divided into functional and nonfunctional. Only 10–15 % of the total number of parathyroid cysts is functional. In addition, the cause of primary

hyperparathyroidism in 1–2 % of cases is a functional parathyroid cyst [6].

Papavramidis T.S. et al. analyzed 359 reports of treatment of parathyroid cysts [5]. The authors reported that the main symptoms of parathyroid cysts are the presence of a mass on the neck in 41.7 % of cases, compression syndrome of the neck in 20.6 %, and dysfunction of the parathyroid glands in the form of hyperparathyroidism in 17.5 %. The authors also report that cysts can be located on the neck from the angle of the mandible to the mediastinum. Thus, the most common localization was the left lobe of the thyroid gland in 31.6 % of cases, the upper mediastinum in 19.3 %, the right lobe of the thyroid gland in 19 %, and the anterior neck in 12.8 %.

The diagnosis of parathyroid cysts is a difficult task. Ultrasound (US) is an ineffective method for the differential diagnosis of cysts of thyroid and parathyroid origin [7]. In addition, scintigraphy has a low sensitivity in relation to detecting an increase in the functional activity of hormonally active cysts in comparison with adenomas of the parathyroid glands [8]. Therefore, the main method for detecting functional cysts of the parathyroid glands is a fine-needle

* Corresponding author at: Mikrorayon Yubileyniy 119–11, Irkutsk, Irkutsk Region, 664049, Russian Federation.

E-mail addresses: lena.isi@mail.ru (E.A. Ilyicheva), gbersenev17@gmail.com (G.A. Bersenev).



Fig. 1. Multi-slice CT scan with angiography: MSCT of the neck organs. 1a - frontal projection, 1b - sagittal projection, 1c - axial projection.

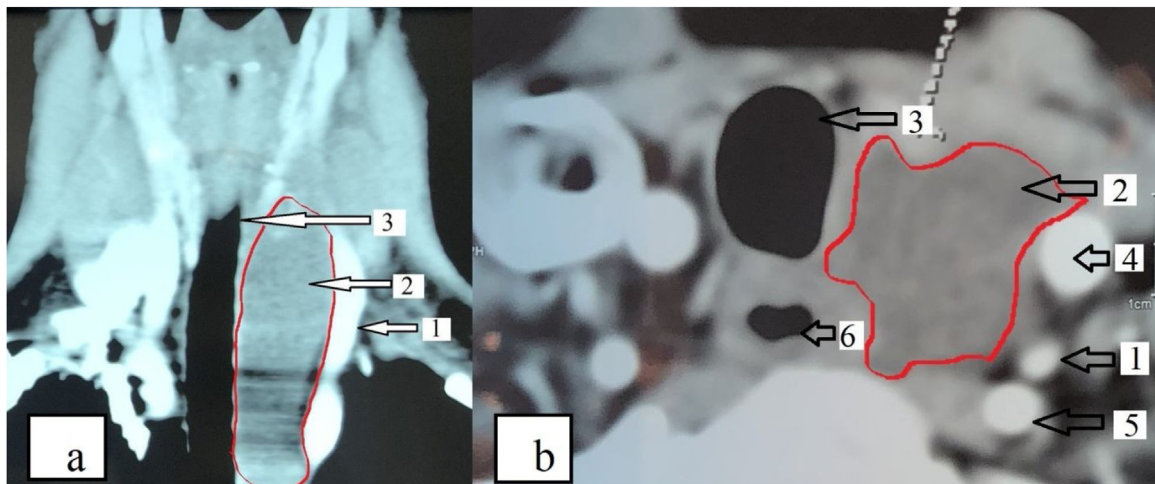


Fig. 2. Multi-slice CT scan with angiography: 2a - Sagittal scan, 2b - Axial scan. 1 - left common carotid artery; 2 - cyst of the left lower parathyroid gland; 3 - trachea; 4 - left internal jugular vein; 5 - left subclavian artery; 6 - esophagus.

aspiration (FNA) of the cyst with the determination of the level of parathyroid hormone [1].

We present our experience of surgical treatment of functional parathyroid cyst in the patient with primary hyperparathyroidism in State Budgetary Institution of Public. This case report has been reported in accordance with the SCARE Criteria [9].

2. Presentation of case

A 57-year-old pensioner Eurasian woman has been under the care of endocrinologist with multinodular goiter for 3 years. In January 2020, the patient was found to have an increase in the level of parathyroid hormone – 128.1 pg/mL (reference values 15.0–68.3 pg/mL). Multispiral computed tomography (MSCT) (February 2020) scan showed the presence of a hypodense fluid formation measuring 110 × 57 × 45 mm in the left lobe of the thyroid gland, which displaces the esophagus and trachea to the right. Due to compression by this formation, the lumen of the trachea is narrowed to 3 mm (Fig. 1).

In March 2020, the patient began to worry about the presence of a mass in the left lateral region of the neck, the appearance of shortness of breath during exercise and the presence of kidney stones. In fact, she has not been suffering from pain in bones and joints and has not had pathological fractures, gastropathy, as well as vitamin D deficiency. The patient has not been using drug, and also she denied the presence of family and hereditary diseases. She had not past surgical history and any other chronic diseases.

Only in august 2020 she was referred to an endocrine surgeon in our clinic to evaluate complaints of shortness of breath, severe dyspnea at rest, a presence of a mass of the neck and persistently elevated level of calcium. In the biochemical analysis of the patient's blood, the serum total calcium level was increased – 2.73 mmol/l (reference values 2.1–2.6 mmol/l), ionized calcium – 1.53 mmol/l (reference values 1.15–1.27 mmol/l), parathyroid hormone – 149.6 pg/ml (reference values 15.0–68.3 pg/ml). The daily urinary excretion of calcium was 2.96 mmol/day (reference values 2.5–6.25 mmol/day). An US scan showed the presence of an anechoic formation of irregular shape with dimensions 81 × 52 × 35 mm, which was located between the lower pole of the left lobe of the thyroid gland and the left neurovascular bundle of the neck, leaving retrosternal. There was not any mass that could be identified as a parathyroid adenoma or parathyroid hyperplasia. The total volume of the thyroid gland was 11.4 cm³, the right lobe was 4.0 cm³, the left lobe was 7.4 cm³. A scintigraphy scan did not show an increase in the functional activity of the parathyroid glands.

Given the pronounced dyspnea and narrowing of the tracheal lumen to 3 mm in diameter, the first stage of the patient underwent FNA of fluid from the cyst (August 27, 2020) in order to decompress the trachea. Evacuated 150 ml of clear liquid. Repeated MSCT (September 8, 2020) scan showed the presence of a fluid formation measuring 80 × 40 × 33 mm, which is placed from the lower edge of the lower pole of the thyroid gland to the level of the aortic arch displacing the trachea medially to the right (Fig. 2).

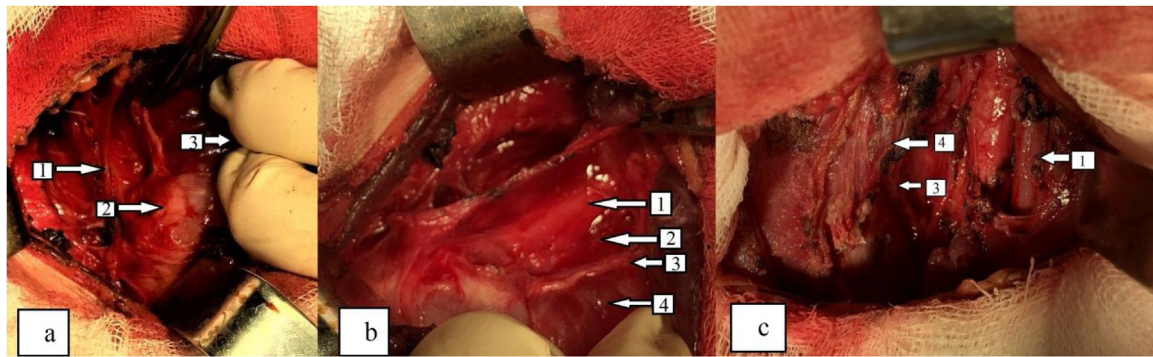


Fig. 3. Intraoperative photography. Arrows indicate anatomical structures. 3a - before cyst aspiration: 1- left common carotid artery; 2 - anterior surface of the cyst of the left lower parathyroid gland; 3- the left lobe of the thyroid gland is abducted. 3b - after cyst aspiration: 1- left common carotid artery; 2- the posterior surface of the cyst of the left lower parathyroid gland; 3- left recurrent laryngeal nerve; 4- the left lobe of the thyroid gland. 3c - after removal of the cyst: 1- left common carotid artery; 3- left recurrent laryngeal nerve; 4- the left lobe of the thyroid gland.

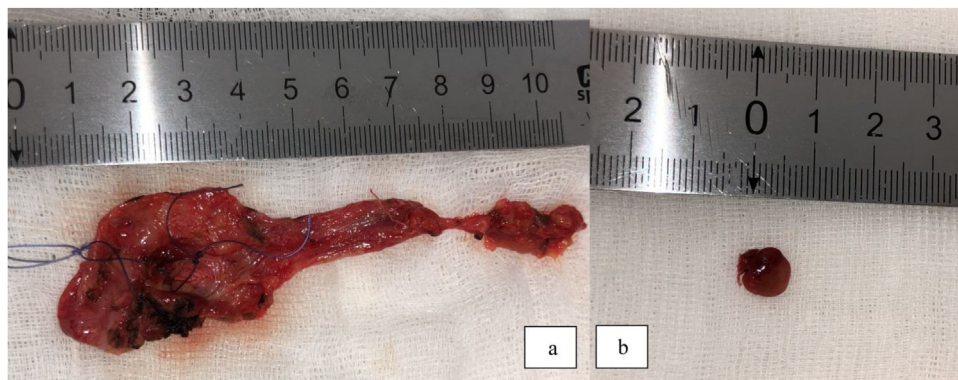


Fig. 4. Macrophotography of operative material. 4a - removed cyst of the left lower parathyroid gland. 4b - removed left superior parathyroid gland.

The fluid formation does not accumulate contrast agent. The parathyroid glands are not reliably visualized. To determine whether the neck cyst was associated with the parathyroid glands, thyroid gland, or neither, FNA of the cyst fluid was performed. FNA revealed the clear color of the cystic fluid in which intact PTH (iPTH) level was markedly higher (>2500 pg/mL) than serum iPTH level. The patient was thus diagnosed with primary hyperparathyroidism caused by a functional parathyroid cyst.

According to the preoperative study and the absence of clinical recovery after FNA of the cyst, surgical intervention was planned in the amount of cervicotomy, intraoperative aspiration of the parathyroid cyst with the determination of the level of iPTH in the cyst fluid, left lower parathyroidectomy with parathyroid cystectomy and intraoperative monitoring of iPTH.

The operation took place on September 14, 2020. The operation was performed by an endocrine surgeon who has 15 years of experience in this area.

A cervicotomy was performed according to a standard technique. A fluid formation (thin-walled cyst) was found ventrally to the left recurrent laryngeal nerve. The cyst was located from the left lower parathyroid gland and occupied the entire space between the left lobe of the thyroid gland, trachea, esophagus medially and the left neurovascular bundle laterally. The upper edge of the cyst was located above the upper pole of the left lobe of the thyroid gland, the lower edge extends along the trachea retrosternally into the upper mediastinum. The left lower parathyroid gland was located on the anterior surface of the cyst with signs of sclerosis, dimensions $0.8 \times 0.5 \times 0.5$ mm, dark brown. The left recurrent laryngeal nerve was intimately attached to the posterior surface of the cyst (Fig. 3). The upper left parathyroid gland $1.0 \times 0.8 \times 0.6$ cm in size,

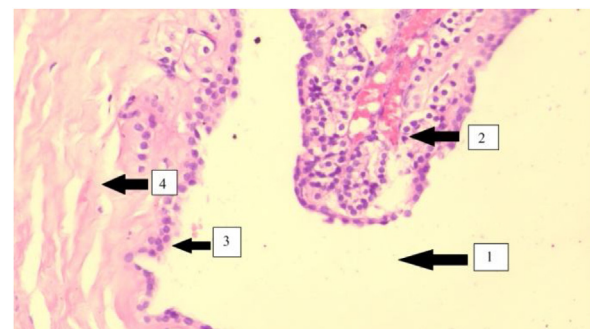


Fig. 5. Microphotography of operative material. Tinted with hematoxylin and eosine. Magnification 10×0.25 . 1- lumen of the cyst of the left lower parathyroid gland; 2 - hyperplastic area of the left lower parathyroid gland; 3- the epithelium of the cyst wall from the dark main cells of the parathyroid gland; 4- fibrous tissue.

dark brown, was found dorsally to the left recurrent laryngeal nerve and cranially to the upper pole of the left lobe of the thyroid gland. FNA of a thin-walled cyst was performed. The level of iPTH in cyst fluid was more than 2500 mg/mL.

The mobilization and removal of the cyst of the left lower parathyroid gland was performed (Fig. 4a). Given the increased size of the left upper parathyroid gland, the operation was continued by its mobilization and removal (Fig. 4b).

The dynamics of the level of intraoperative monitoring of iPTH was as follows: before the skin incision – 128.5 pg/mL, at the time of mobilization of the cyst of the left lower parathyroid gland – 197.1 pg/mL, 10 min after removal of the cyst of the left lower

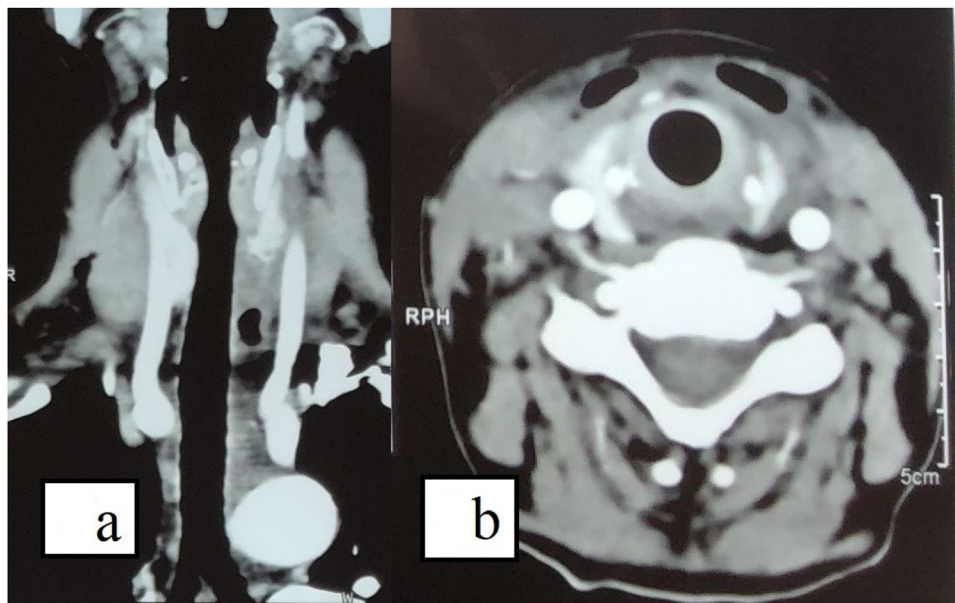


Fig. 6. Multi-slice CT scan with angiography: 6a – Frontal scan, 6b – Axial scan.

parathyroid gland – 125.3 pg/mL, after 10 min after removal of the left upper parathyroid gland – 124.5 pg/mL.

According to the histological study, the cyst wall was presented by fibrous tissue, which was lined with epithelium from the dark main cells of the parathyroid gland. In the section of the cyst wall, tissue of the parathyroid gland with signs of hyperplasia (left lower parathyroid gland) was found. The removed left upper parathyroid gland was represented by the parathyroid tissue of the usual structure (Fig. 5).

In the postoperative period, laryngoscopy was performed, on which the normal mobility of the vocal folds was established. On the first day after surgery, the level of iPTH was 77,2 pg/ml, the level of serum total calcium was 2,89 mmol/l. On the 4th day, MSCT was performed in order to search for additional parathyroid glands. In the projection of the bed of the removed cyst, gas is visualized, the fatty tissue is infiltrated. The parathyroid glands are not reliably visualized (Fig. 6).

The patient was discharged on the 7th day after the operation under the care of an outpatient surgeon and endocrinologist. At the time of discharge, the level of serum total calcium was 2.6 mmol/l. The patient was examined by the operating surgeon 2 months after the operation. According to the patient, she feels great and does not actively complain. She breathes freely and independently, allows herself light anaerobic loads (light jogging, yoga). In the patient's opinion, the treatment carried out helped her. In the control biochemical analysis of the patient's blood (November 16, 2020), the serum total calcium level was increased – 2,35 mmol/l, ionized calcium – 1.23 mmol/l, parathyroid hormone – 45.4 pg/mL.

3. Discussion

This clinical case confirms the existing point of view that parathyroid cysts are a rare cause of hyperparathyroidism and a difficult diagnostic task [1]. In our patient, the parathyroid cyst was manifested by the presence of a mass on the left lateral surface of the neck, an increase in the level of parathyroid hormone, and hypercalcemia. In addition, the cyst was large and displaced the trachea in the opposite direction with the development of neck compression syndrome, which was manifested by shortness of breath during exercise. According to the data of preoperative examination (ultrasound, MSCT), we found the presence of large fluid

formation near the left lobe of the thyroid gland and the absence of enlarged parathyroid glands. By scintigraphy, the localization of enlarged parathyroid glands was not established. This confirms literature data that the sensitivity of imaging methods decreases when parathyroid cysts are detected [7].

This clinical case showed that the main role in the diagnosis was played by fine-needle aspiration of the cyst fluid and an increased level of iPTH in the cyst fluid. Based on the experience of surgical treatment of parathyroid cysts, operation was planned with intraoperative fine-needle aspiration of the parathyroid cyst with determination of the level of parathyroid hormone in the cyst fluid, left lower parathyroidectomy with cystectomy and intraoperative monitoring of iPTH. In our observation, intraoperative monitoring of iPTH had low sensitivity in detecting and assessing the radicality of surgery.

4. Conclusion

Parathyroid cysts in primary hyperparathyroidism are a difficult diagnostic and therapeutic task. A comprehensive and thorough assessment of all methods of visualization of the parathyroid glands at the preoperative stage, including the use of MSCT angiography and FNA under ultrasound control, is required in order to establish a diagnosis. Functional parathyroid cysts require surgical treatment. Intraoperative monitoring of iPTH has low sensitivity in relation to the detection and assessment of the radicality of the operation for parathyroid cysts.

Declaration of Competing Interest

The authors report no declarations of interest.

Sources of funding

No funds were received for any part of this case report.

Ethical approval

This case report is exempt from ethical approval at our institution as this is not a research study.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Author contribution

Ilyicheva E.A. – the surgical stage of therapy; data analysis and interpretation; development of the concept and design; substantiation of the manuscript and verification of critical intellectual content; editing and final approval of the manuscript.

Bersenev G.A. – development of the concept and design; collection of material, analysis and interpretation of data, substantiation of the manuscript and verification of critical intellectual content.

Registration of research studies

Not applicable.

Guarantor

Ilyicheva E.A.

Provenance and peer review

Not commissioned, externally peer-reviewed.

Acknowledgments

We are grateful to the following persons for their role during this study: Valery Makhutov, Chief of Surgical Department, State Budgetary Institution of Public Health “Irkutsk Regional Clinical

Hospital”, Irkutsk (physician-in-chief P.E. Dudin) for the support during all stages of therapy and assistance in manuscript drafting; Tatyana Boyko and Ekaterina Pisarskaya, radiologists, State Budgetary Institution of Public Health “Irkutsk Regional Clinical Hospital”, Irkutsk (physician-in-chief P.E. Dudin) in the preparation for publishing (Figs. 1, 2 and 6), respectively; Elena Rozhanskaya, anatomic pathologist, Oleg Kanya, Chief of Anatomic Pathology Department, State Budgetary Institution of Public Health “Irkutsk Regional Bureau of Anatomic Pathology” (Director Oleg Kanya) in the preparation for publishing (Fig. 5).

References

- [1] G. Ippolito, F.F. Palazzo, F. Sebag, M. Sierra, C. De Micco, J.F. Henry, A single-institution 25-year review of true parathyroid cysts, *Langenbecks Arch. Surg.* 391 (2006) 13–18, <http://dx.doi.org/10.1007/s00423-005-0579-y>.
- [2] C. Cappelli, M. Rotondi, I. Piroola, E. De Martino, P. Leporati, F. Magri, E. Agabiti Rosei, L. Chiovato, M. Castellano, Prevalence of parathyroid cysts by neck ultrasound scan in unselected patients, *J. Endocrinol. Invest.* 32 (2009) 357–359, <http://dx.doi.org/10.1007/BF03345727>.
- [3] I. Sandström, On a new gland in man and several mammals, *Upsala Lak Foren Forh* (1879).
- [4] D. Goris, Extirpation of three cystic parathyroid lobules, *Ann. Soc. Belge Chir.* 5 (1905) 394.
- [5] T.S. Papavramidis, A. Chorti, I. Pliakos, S. Panidis, A. Michalopoulos, Parathyroid cysts a review of 359 patients reported in the international literature, *Med. (United States)* 97 (2018), <http://dx.doi.org/10.1097/MD.00000000000011399>.
- [6] J. Rosenberg, R. Orlando, M. Ludwig, L.J. Pyrtek, Parathyroid cysts, *Am. J. Surg.* 143 (1982) 473–480, [http://dx.doi.org/10.1016/0002-9610\(82\)90198-2](http://dx.doi.org/10.1016/0002-9610(82)90198-2).
- [7] K. Suzuki, A. Sakuta, C. Aoki, Y. Aso, Hyperparathyroidism caused by a functional parathyroid cyst, *BMJ Case Rep.* 2013 (2013), <http://dx.doi.org/10.1136/bcr-2012-008290>.
- [8] N.A. Johnson, L. Yip, M.E. Tublin, Cystic parathyroid adenoma: sonographic features and correlation with 99mTc-sestamibi SPECT findings, *Am. J. Roentgenol.* 195 (2010) 1385–1390, <http://dx.doi.org/10.2214/AJR.10.4472>.
- [9] R.A. Agha, M.R. Borrelli, R. Farwana, K. Koshy, A. Fowler, D.P. Orgill, For the SCARE Group, The SCARE 2018 statement: updating consensus Surgical Case Report (SCARE) guidelines, *Int. J. Surg.* 60 (2018) 132–136, <http://dx.doi.org/10.1016/j.ijssu.2018.10.028>.

Open Access

This article is published Open Access at [sciencedirect.com](https://www.sciencedirect.com). It is distributed under the [IJSCR Supplemental terms and conditions](#), which permits unrestricted non commercial use, distribution, and reproduction in any medium, provided the original authors and source are credited.