ARCHIVES OF HEAD AND NECK SURGERY
ISSN 2595-2544

CASE REPORT

Supernumerary ectopic parathyroid located in the left carotid sheath

Ana Karenina Nobre Fonseca de Souza[1], Liliane Queiroz de Lira[2], Rodrigo Azevedo de Oliveira[3]

Abstract

A 34 years old male patient with chronic renal failure and history of total parathyroidectomy with parathyroid autotransplantation evolved with persistence of elevated PTH, bone pain. Cervical ultrasonography and parathyroid scintigraphy with sestaMIBI revealed changes in the topography of the left submandibular gland. He underwent cervical exploration showing supranumerary ectopic parathyroid, found in the left carotid sheath at I/IIA levels, with histopathological confirmation by frozen section biopsy. On the first postoperative day, serum PTH decreased by more than 95%, thus, confirming operative success. The symptoms improved along the follow up.

Keywords: parathyroidectomy; chronic kidney disease; reoperation.

[1]Universidade Federal do Rio Grande do Norte (UFRN), Hospital Universitário Onofre Lopes, Departamento de Cirurgia de Cabeça e Pescoço, Natal, RN, Brasil

[2]Universidade Federal do Rio Grande do Norte (UFRN), Hospital Universitário Onofre Lopes, Departamento de Otorrinolaringologia, Natal, RN, Brasil

[3]Universidade Federal do Rio Grande do Norte (UFRN), Hospital Universitário Onofre Lopes, Departamento de Nefrologia, Natal, RN, Brasil

Financial support: None.
Conflicts of interest: No conflicts of interest declared concerning the publication of this article.
Submitted: September 08, 2018.
Accepted: October 25, 2018.

The study was carried out at Unidade de Cabeça e Pescoço, Hospital Universitário Onofre Lopes, Universidade Federal do Rio Grande do Norte (UFRN), Natal, RN, Brasil.

Introduction

Secondary hyperparathyroidism (SHPT) is characterized by hypersecretion of the parathyroid glands in response to the imbalance of mineral homeostasis, especially Calcium, and can be associated with chronic kidney disease (CKD)[1]. Some patients who have CKD and who, while waiting for renal transplantation, undergo hemodialysis for several years, develop SHPT refractory to clinical therapy, requiring surgical treatment that consist of total parathyroidectomy (PTX) with autotransplantation. This procedure has excellent effects on symptomatology as well as in reducing the patients' mortality rates[1].

During postoperative follow-up, high PTH levels suggest the presence of supernumerary and/or ectopic parathyroid glands, which is the main cause of persistent disease after surgery[2].

In this report, we present the case of a patient with CKD and a history of PTX with heterotopic autotransplantation who developed a significant PTH rate after surgery.

Case report

A 34 years old male patient presented CKD for 10 years due to recurrent urinary tract infection. During follow-up at the kidney pre-transplant outpatient clinic, he presented bone pain refractory to clinical treatment, facial deformity,

loss of 6 cm in stature and difficulty to walk around requiring wheelchair use, as well as PTH of 2892.8 pg/mL, when he underwent PTX excision of the four parathyroid glands and heterotopic autotransplantation in upper right limb. Two years later, the postoperative evaluation revealed persistence of elevated PTH rates (2718 pg/ml), maintenance of bone deformity and bone pain that required the use of opioids for symptomatic control. Thus, the possibility of a supranumerary ectopic parathyroid gland was suggested. The cervical ultrasonography showed an oval, well delimited, hypoechogenic nodule with central and peripheral vascularization in the left submandibular region. The parathyroid scintigraphy by sestaMIBI (MIBI) revealed increased radiopharmaceutical uptake in the left submandibular gland topography in relation to the contralateral gland. The patient was referred to a new surgical approach. He underwent a cervical exploration with parathyroid findings in the carotid sheath I/IIA (Figure 1), at left, with histopathological confirmation of the parathyroid gland by means of a freezing biopsy (Figure 2). Our department does not have intraoperative PTH in order to quantify the serum value of this hormone during the procedure. The PTH serum level in the first postoperative day was 133 pg/mL, demonstrating a reduction above 95% of the initial value, to the detriment of the implant in the upper limb. This result confirms the operative success. The patient continues in outpatient follow-up with improvement of the symptomatology.

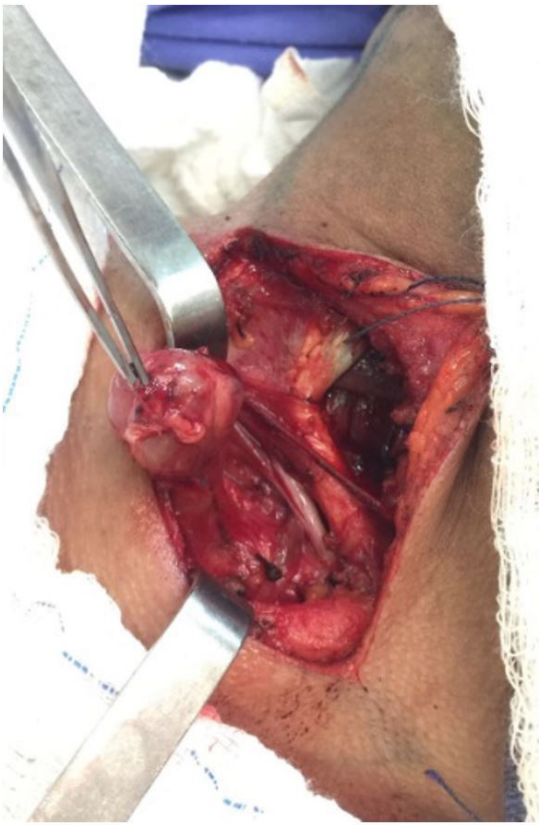

Figure 1. Ectopic supernumerary parathyroid in carotid sheath level I/IIA at left.

Figure 2. Product of resection whose analysis by biopsy of freezing has confirmed to be parathyroid gland.

Discussion

The presence of ectopic and supernumerary parathyroid glands in patients submitted to PTX due to hyperparathyroidism associated with CKD is significant[3]. It is believed that abnormal migration of the parathyroid glands occurs during embryogenesis, when the upper glands migrate with the thyroid and remain close to their lobes, while the lower parathyroid glands move with the thymus, traveling long distances, which justifies the heterogeneous pattern of localization in relation to the superior ones[4]. A rate of 13.6% of ectopic parathyroid glands and 4.8% of supernumerary parathyroid glands has been reported[5].

This fact justifies the use of preoperative complementary examinations as a method of surgical planning, since they present low sensitivity for the diagnosis of ectopic/supernumerary glands. It is worth mentioning that careful intraoperative search should also be considered. The rapid intraoperative PTH is a valuable tool in these cases, since the persistence of elevated PTH or even an insufficient decline in its value suggests a supernumerary gland, indicating a larger extensive cervical exploration, especially in the main sites of ectopic glands, as well as: in the case of upper parathyroid glands, the retroesophageal position and, in the case of the lower parathyroid glands, the thymic region[2,5].

Thus, we conclude that PTX should be the first choice of surgical treatment for advanced SHPT refractory to clinical treatment, and that surgical planning and postoperative follow-up should always consider possible anatomical variations, since the remaining glandular tissue may evolve with hyperfunction and autonomy leading to high morbidity and mortality, requiring a new surgical approach during long-term care[3,4].

***Correspondence**
Liliane Queiroz de Lira
Universidade Federal do Rio Grande do Norte (UFRN), Hospital Universitário Onofre Lopes, Departamento de Otorrinolaringologia
Avenida Nilo Peçanha, 620, Petrópolis
CEP 59012-300, Natal (RN), Brasil
Tel.: +55 (xx) (84) 3342-5041
E-mail: lilianelira_@hotmail.com

Authors information
AKNFS - Medical at the Universidade Federal do Rio Grande do Norte (UFRN), Hospital Universitário Onofre Lopes, Departamento de Cirurgia de Cabeça e Pescoço. LQL - Medical resident at the Universidade Federal do Rio Grande do Norte (UFRN), Hospital Universitário Onofre Lopes, Departamento de Otorrinolaringologia. RAO - Medical at the Universidade Federal do Rio Grande do Norte (UFRN), Hospital Universitário Onofre Lopes, Departamento de Nefrologia.

References

1. Magnabosco FF, Tavares MR, Montenegro FLM. Tratamento cirúrgico do hiperparatireoidismo secundário: revisão sistematizada da literatura. Arq Bras Endocrinol Metabol. 2014;58(1):1-8. PMid:24728158.

2. Gomes EM, Nunes RC, Lacativa PG, Almeida MH, Franco FM, Leal CT, Patrício PJ Fo, Farias ML, Gonçalves MD. Localização de paratireóides ectópicas e extranumerárias em pacientes com hiperparatireoidismo secundário à insuficiência renal crônica. Acta Cir Bras. 2007;22(2):105-9. **http://dx.doi.org/10.1590/S0102-86502007000200005**. PMid:17375215.

3. Tominaga Y, Katayama A, Sato T, Matsuoka S, Goto N, Haba T, Hibi Y, Numano M, Ichimori T, Uchida K. Re-operation is frequently required when parathyroid glands remain after initial parathyroidectomy for advanced secondary hyperparathyroidism in uraemic patients. Nephrol Dial Transplant. 2003;18(Suppl 3):iii65-70. **http://dx.doi.org/10.1093/ndt/gfg1017**. PMid:12771305.

4. Hojaij F, Vanderlei F, Plopper C, Rodrigues CJ, Jácomo A, Cernea C, Oliveira L, Marchi L, Brandão L. Parathyroid gland anatomical distribution and relation to anthropometric and demographic parameters: a cadaveric study. Anat Sci Int. 2011;86(4):204-12. PMid:21850415.

5. Andrade JS, Mangussi-Gomes JP, Rocha LA, Ohe MN, Rosano M, das Neves MC, Santos RO. Localization of ectopic and supernumerary parathyroid glands in patients with secondary and tertiary hyperparathyroidism: Surgical description and correlation with preoperative ultrasonography and Tc99m-Sestamibi scintigraphy. Braz J Otorhinolaryngol. 2014;80(1):29-34. PMid:24626889.