



Scientific letters

Non-Functioning Parathyroid Cysts Refractory to Conservative Treatment[☆]



Quistes no funcionantes de paratiroides refractarios al tratamiento conservador

Parathyroid cysts were first described in 1880 by Sandström, but the first surgical resection was not performed until 1905 by Goris. These cysts can be classified as functioning, which occur with primary hyperparathyroidism, or non-functioning. The latter are the most frequent, representing 80%–90% of all parathyroid cysts.^{1,2} In addition, cystic parathyroid lesions account for 0.075% of cervical ultrasounds.³

Functional parathyroid cysts are managed like primary hyperparathyroidism, whose treatment of choice is surgery. Thus, functioning parathyroid cysts account for 3% of parathyroidectomies performed by primary hyperparathyroidism.⁴

In contrast, the management of non-functioning cysts is conservative, with drainage by needle aspiration being the treatment of choice. Regarding the etiopathogenesis of these non-functioning cysts, there are 2 theories: the first is that they originate in the embryonic period from the 3rd and 4th pharyngeal arches; the second indicates that they originate by fusion of parathyroid microcysts caused by degeneration of the gland or retention of secretions that form microcysts.¹

The objective of this case report is to analyze the management of non-functioning parathyroid cysts refractory to drainage with needle aspiration.

Patients with parathyroid cysts that met the following criteria are included in this report: (a) ultrasound or computed tomography (CT) diagnosis of parathyroid cysts; (b) fluid obtained by needle aspiration, with limited cellularity and a concentration of parathyroid hormone (PTH) greater than in plasma (>9–65 pg/ml); (c) no hyperparathyroidism (normal plasma PTH and calcium levels); (d) previous treatment with percutaneous drainage; and (e) recurrence of the cyst, with a size greater than 3 cm in diameter.

Four patients met the inclusion criteria (Table 1), which represents 0.43% of all parathyroid disease treated surgically at our hospital in the same period of time. Excluded from the series were 3 treated parathyroid cysts as they were functioning and treated as primary hyperparathyroidism. 75% (n=3) of patients were male and mean age was 37.7±13.8 years. 75% (n=3) of the non-functioning cysts were located in the left lower parathyroid gland. In the 4 cases (100%), surgery was indicated, entailing parathyroidectomy of the affected gland. Being non-functioning cysts, they did not require intraoperative monitoring of PTH. There were no complications in the postoperative period. After a mean follow-up of 13.2±8.9 years, the patients have not presented recurrence of the disease.

Non-functioning parathyroid cysts are usually asymptomatic,^{2,5} except those that reach a size greater than 3 cm, which tend to cause symptoms due to the compression of neighboring structures.^{1,2} The initial treatment is percutaneous drainage, which is ambulatory and can resolve the condition in up to 50% of cases, or at least maintain it at a size that does not produce compressive symptoms.⁶

In cases of cyst recurrence after aspiration with an acquired size greater than 3 cm, a more definitive treatment is recommended. Some authors propose further needle aspirations, but in most cases there are new cyst recurrences.^{7,8} The most effective treatment is surgery, as it achieves definitive resolution of the cyst. Classically the main reason against surgery was the associated morbidity. Today, however, in experienced endocrine surgery units, this surgery is performed with practically no morbidity,^{9,10} as observed in our series. Remember that, as it is neither tumor tissue nor functioning, the risk of parathyromatosis in case of rupture of the cyst is virtually non-existent.

[☆] Please cite this article as: Ruiz J, Ríos A, Rodríguez JM, Parrilla P. Quistes no funcionantes de paratiroides refractarios al tratamiento conservador. Cir Esp. 2018;96:52–54.

Table 1 – Characteristics of Patients With Recurrent Non-Functioning Parathyroid Cysts.

	Case 1	Case 2	Case 3	Case 4
Age (yrs)	43	55	27	26
Sex	Female	Male	Male	Male
Symptoms	Asymptomatic	Cervical discomfort when swallowing	Asymptomatic	Cervical discomfort when swallowing
Physical examination	Palpable and non-painful left cervical nodule	Palpable and painful left cervical nodule	Palpable and non-painful left cervical nodule	Palpable and painful right cervical nodule
Analysis				
Ca (mg/dL)	9	9.5	9.6	9
P (mg/dL)	3.5	2.3	4.1	3.3
PTH (mg/dL)	24	39	22	11
TSH (IU/mL)	3.66	2.34	1.55	1.88
T4l (ng/dL)	1.24	1.18	1.34	1.60
Ultrasound	4.2 cm cyst in the lower pole of the LTL	4.8 cm cyst in the lower pole of the LTL	4 cm cyst in the lower pole of the LTL	3.4 cm cyst in the lower pole of the RTL
FNA				
Cytology	Clear liquid with limited cellularity	Clear liquid with limited cellularity	Clear liquid with limited cellularity	Clear liquid with limited cellularity
PTH (pg/mL)	2001	370	443	400
No. of previous aspirations	1	3	1	2
Surgery	Yes	Yes	Yes	Yes
Resected gland	LIPG	LIPG	LIPG	RIPG
Complications	No	No	No	No
Histopathology	Parathyroid cyst	Parathyroid cyst	Parathyroid cyst	Parathyroid cyst
Follow-up (yrs)	21	20	10	2
Recurrence	No	No	No	No

RIPG: right inferior parathyroid gland; LIPG: left inferior parathyroid gland; RTL: right thyroid lobe; LTL: left thyroid lobe; FNA: fine-needle aspiration.

In recent years, other non-surgical therapeutic alternatives have been developed and promoted. For instance, radiofrequency has been gaining indications in benign disease, both thyroid and parathyroid, with promising results. However, the experience to recommend its use in parathyroid disease is minimal, especially in cystic lesions where the results are worse than in solid lesions. Sclerosis with tetracycline or ethanol^{7,11} has been recommended by some authors, and has even been recommended as a second therapeutic step after the failure of percutaneous drainage by needle aspiration. However, parathyroid cysts >3 cm, because of their size, are usually close to the recurrent nerve, which is frequently injured. Therefore, our group does not recommend this technique. In any case, we should keep in mind that, given the lack of experience in this disease, recommendations cannot be restrictive.

In conclusion, for the treatment of non-functioning parathyroid cysts refractory to percutaneous needle-aspiration drainage, surgery is a safe and effective therapeutic option in endocrine surgery units.

REFERENCES

- Pontikides N, Karras S, Kaprara A, Cheva A, Doumas A, Botsios D, et al. Diagnostic and therapeutic review of cystic parathyroid lesions. *Hormones (Athens)*. 2012;11:410-8.
- Rossi ED, Revelli L, Giustozzi E, Straccia P, Stigliano E, Lombardi CP, et al. Large non-functioning parathyroid cysts: our institutional experience of a rare entity and a possible pitfall in thyroid cytology. *Cytopathology*. 2015;26:114-21.
- Cappelli C, Rotondi M, Pirola I, de Martino E, Leporati P, Magri F, et al. Prevalence of parathyroid cysts by neck ultrasound scan in unselected patients. *J Endocrinol Invest*. 2009;32:357-9.
- McCoy KL, Yim JH, Zuckerbraun BS, Ogilvie JB, Peel RL, Carty SE. Cystic parathyroid lesions functional and nonfunctional parathyroid cysts. *Arch Surg*. 2009;144:52-6.
- Ippolito G, Palazzo FF, Sebag F, Sierra M, de Micco C, Henry JF. A single-institution 25 year review of true parathyroid cysts. *Langenbecks Arch Surg*. 2006;391:13-8.
- Prinz RA, Peters JR, Kane JM, Wood J. Needle aspiration of nonfunctioning parathyroid cysts. *Am Surg*. 1990;56:420-2.
- Sung JY, Baek JH, Kim KS, Lee D, Ha EJ, Lee JH. Symptomatic nonfunctioning parathyroid cysts role of simple aspiration and ethanol ablation. *Eur J Radiol*. 2013;82:316-20.
- Ujiki MB, Nayar R, Sturgeon C, Angelos P. Parathyroid cyst often mistaken for a thyroid cyst. *World J Surg*. 2007;31:60-4.
- Zambudio AR, Rodríguez J, Riquelme J, Soria T, Canteras M, Parrilla P. Prospective study of postoperative complications after total thyroidectomy for multinodular goiters by surgeons with experience in endocrine surgery. *Ann Surg*. 2004;240:18-25.
- Amondarain JA, Aribé X, Olalla C, Echenique M. Quiste paratiroides e hiperparatiroidismo. *Cir Esp*. 2008;83:97-8.
- Kim JH. Ultrasound guided sclerotherapy for benign non thyroid cystic mass in the neck. *Ultrasonography*. 2014;33:83-90.

José Ruiz^{ab}, Antonio Ríos^{ac*}, José Manuel Rodríguez^{ac},
Pascual Parrilla^{ac}

^aDepartamento de Cirugía, Ginecología Obstetricia y Pediatría,
Universidad de Murcia, Murcia, Spain

^bServicio de Cirugía General y de Aparato Digestivo,
Hospital Clínico Universitario Virgen de la Arrixaca,
El Palmar, Servicio Murciano de Salud,
Murcia, Spain

^cInstituto Murciano de Investigación Bio-Sanitaria Virgen de la
Arrixaca (IMIB-Arrixaca), Murcia, Spain

*Corresponding author.

E-mail address: arzrios@um.es (A. Ríos).

2173-5077/

© 2017 AEC. Published by Elsevier España, S.L.U. All rights reserved.

Spontaneous Dissection of the Superior Mesenteric Artery[☆]



Disección espontánea de arteria mesentérica superior

Spontaneous dissection of the superior mesenteric artery (SMA) is a rare disease, with an incidence of 0.06%.¹ The SMA is the most common location of isolated dissection and is the second peripheral artery in frequency after the internal carotid artery. The first case was described by Bauersfeld in 1947,² and the review of the literature has identified only 168 cases since then.³ It is more frequent in males (4:1) in the fifth decade of life.⁴ When there is suspicion of abdominal vascular compromise, CT scan is recommended as the initial diagnostic technique, while arteriography is used in patients with worsening symptoms. Anticoagulant treatment is currently the basis of conservative therapy for SMA dissection and is associated with increasingly better results.

We present the case of a 58-year-old male patient with a history of HTN, active smoking and dyslipidemia, who reported very intense abdominal pain over the previous 4 h (EVA 10/10). The patient was hemodynamically stable and showed no signs of peritoneal irritation. Lab workup demonstrated leukocytosis and elevated PCR, with no metabolic acidosis.

Abdominal CT scan showed a filling defect in the proximal segment of the SMA, suggestive of a thrombus that led to stenosis of approximately 60%. There was also a linear defect inside the lumen of the vessel that extended to a jejunal branch, which may have corresponded with a flap associated with the dissection. Adequate distal filling was observed with no signs of intestinal ischemia.

Initially, we opted for a conservative approach with anti-coagulant therapy (heparin). The progression of the patient's condition became torpid, with more pain and guarding in the right hemiabdomen. The leukocytosis continued, associated with neutrophilia and mild metabolic acidosis. Given the worsening symptoms, we decided to perform CT angiography, which showed evidence of a small repletion defect at the origin of the ileocolic artery, with distal opacification related with a

partially occlusive thrombus. The ileal loops located in the iliac fossa and right flank presented intestinal suffering (Fig. 1). Given the hemodynamic stability, we decided to perform a radiological approach using arteriography of the SMA, where a dissection flap was observed within the first few centimeters, causing obstruction of all the ileal branches, with no evidence of stenosis at the origin (Fig. 2).

Endovascular treatment was not considered indicated by the interventional radiology department because of the risk for aggravating the vascular obstruction.

Urgent laparotomy confirmed ischemia of the jejunum, ileum, ascending and transverse colon. Damage control surgery was performed, entailing the resection of 150 cm of small intestine and extended right hemicolectomy with no anastomosis or stoma. Revision surgery 48 h later involved ileocolic anastomosis in a second phase. The patient's condition progressed satisfactorily, and anticoagulation was maintained for 6 months.

The pathogenesis of spontaneous SMA dissection is uncertain, although some risk factors have been identified, such as cystic medial degeneration, fibromuscular dysplasia, connective tissue diseases, arteriosclerosis, hypertension, abdominal aortic aneurysm and trauma, although it generally occurs in previously healthy patients.⁵

The mean distance from the SMA ostium to the beginning of the dissection is from 1.5 to 3 cm because it is a transition zone between the fixed retropancreatic proximal part of the artery and the relatively mobile more distal part that pivots with the movements of the bowel. It is at this point that an intimal flap is normally formed, which allows blood to enter the interior of the medial layer, causing a longitudinal dissection along the laminar plane of the vessel.⁶

The most frequent clinical presentation is acute abdominal pain due to intestinal ischemia, intraperitoneal hemorrhage due to rupture of the SMA, or the inflammatory response

[☆] Please cite this article as: Abascal Amo A, Rodríguez Sánchez A, García Sanz Í, Gimeno Calvo FA, Martín Álvarez JL. Disección espontánea de arteria mesentérica superior. Cir Esp. 2018;96:54–56.