Urology Case Reports 33 (2020) 101400

Contents lists available at ScienceDirect

Urology Case Reports

journal homepage: http://www.elsevier.com/locate/eucr

Endourology

A case of adrenal lymphangioma resected laparoscopically with minimal invasiveness

Hiroshi Yaegashi *, Takahiro Nohara, Kazuyoshi Shigehara, Kouji Izumi, Yoshifumi Kadono, Atsushi Mizokami

Department of Integrative Cancer Therapy and Urology, Kanazawa University Graduate School of Medical Science, Kanazawa, Ishikawa, Japan

ARTICLE INFO

Keywords:
Adrenal cystic tumor
Lymphangioma
Catecholamine
MIBG
Pheochromocytoma

ABSTRACT

A 33-year-old female presented to us with a left adrenal cystic tumor with a maximum diameter of 70 mm. Because malignant tumor and pheochromocytoma could not be excluded, she underwent left laparoscopic transperitoneal adrenalectomy. The cystic tumor was stored to an endoscopically inactive treatment device and was subsequently punctured within the device; thus, tumor removal could be performed with minimum incision. Pathological findings showed highly suggestive of a cystic lymphangioma. The punctate was found to have an extremely high catecholamine titer. To avoid unnecessarily exposing the tumor contents, the treatment approach described in this report is reasonable and worth reporting.

Introduction

Owing to advances in diagnostic imaging, the cases of incidentally encountering adrenal masses have been increasing. Interestingly, although, to date, there is a relatively consistent opinion on the surgical indication for adrenal solid lesions, the differential diagnosis and treatment indications for asymptomatic and inactive functional adrenal cystic masses have been not been fully established.

Adrenal cystic tumors are a rare occurrence, and the contents of adrenal cysts often show high adrenocortical hormone or catecholamine titers.[1] Therefore, cyst rupture involves the risk of exposure to these catecholamines.

Here we present the case of a patient with an adrenal lymphangioma in whom the tumor was removed by puncturing it within an endoscopically inactive treatment device.

Case presentation

A 33-year-old unmarried female visited a hospital with the complaint of abdominal pain in January 2018. Contrast-enhanced abdominal computed tomography scan showed the presence of a left adrenal mass with a maximum diameter of 70 mm (Fig. 1A). She was then referred to our institute, where she underwent a plain T2-weighted abdominal magnetic resonance imaging; it indicated the presence of a left adrenal cystic tumor (Fig. 1B). 131I-meta-iodobenzylguanidine (MIBG) scintigraphy showed no abnormal uptake in the left adrenal gland (Fig. 1C). Both blood test and urinalysis showed no abnormal findings; however, a non-significant elevation in urinary adrenaline was observed following urinary catecholamine examination. Because both a malignant tumor and MIBG-negative pheochromocytoma could not be excluded completely, the patient underwent left laparoscopic transperitoneal adrenalectomy in June 2018.

Laparoscopy was performed using the transperitoneal lateral decubitus approach for best exposure of the gland and vessels. The Hasson technique was used to create pneumoperitoneum, and the operation was performed using four working trocars. After dividing the splenocolic ligament and opening the Gerota's fascia, the left adrenal cystic tumor was adequately exposed. The adrenal cystic tumor was then carefully peeled, and the adrenal central vein was ligated using a sealing device and divided. As a result of the prudent operation, the left adrenal gland was extracted without rupture. The cystic tumor was stored in an endoscopically inactive treatment device (Endo Catch II TM, Medtronic, Minnesota, US) and was punctured and aspirated with a syringe within the device using a 23G needle under dual guiding of laparoscope and direct observation from the body surface to avoid to puncturing the bag itself. Thus, tumor removal was performed with minimum incision of 2.5

Abbreviations: MIBG, meta-iodobenzyl guanidine.

* Corresponding author.
E-mail addresses: hiroshining@hotmail.com (H. Yaegashi), t_nohara704@yahoo.co.jp (T. Nohara), kshigehara0415@yahoo.co.jp (K. Shigehara), azuizu2003@yahoo.co.jp (K. Izumi), yskadono@yahoo.co.jp (Y. Kadono), mizokami@staff.kanazawa-u.ac.jp (A. Mizokami).

https://doi.org/10.1016/j.eucr.2020.101400
Received 6 August 2020; Received in revised form 29 August 2020; Accepted 1 September 2020
Available online 2 September 2020

cm.

The operation was uneventful, and the patient was discharged after 8 days without complications. Histopathological examination of the resected specimen highly suggested a lymphangioma, and immunohistochemistry staining of the cells lining the cystic spaces were positive for CD34 and D2-40 (Fig. 2A and B). Cytological examination results of the cyst contents were negative for malignancy. However, surprisingly, catecholamine titers of the cyst contents were markedly increased (Table 1). Notably, the patient stopped complaining of the mild headache that she occasionally used to feel preoperatively.

Discussion

According to a summary of 53 cases of adrenal lymphangioma, the tumor more commonly affects individuals aged between 16 and 60 years (mean 39.5 years) with a predominance for female sex (12 males and 36 females in the case summary).[2] The right side is significantly more commonly affected (27 cases affected on the right and 17 on the left side). The tumor size ranged between 2.0 and 35.0 cm, and the symptoms depended on the size and location of the mass. Specifically, patients presented with abdominal pain, hypertension, palpable mass, partial small-bowel obstruction, unrelated or no complaints, etc.[2] In the present case, the tumor may have been the cause of the abdominal pain reported by the patient at her first visit.

Adrenal cyst contents often show high adrenocortical hormone or catecholamine titers; the spread of the tumor from the adrenal tissue into the cyst is presumed to be the underlying cause for this.[1] The patient complained of headache preoperatively, which disappeared postoperatively. We believe that catecholamines may have been leaking into the blood following some kind of stimulation and causing the headache.

Adrenal solid tumors are relatively easy to distinguish on diagnostic imaging, whereas adrenal cystic tumors generally exhibit poor characteristic diagnostic imaging findings with different presentations, making the diagnosis difficult.[3] In the present case, a malignant tumor could not be excluded based on the tumor size. Additionally, despite of its negativity for MIBG, pheochromocytoma with cystic degeneration could not be ruled out.

The reason why we could not be ruled out cystic pheochromocytoma is that it is reported that patients of this disease often have asymptomatic or chronic abdominal pain and no abnormalities are observed in laboratory findings in a cohort.[4] Considering these facts, it was still difficult to exclude cystic pheochromocytoma preoperatively even in this case.

Currently, surgical resection is the only treatment for adrenal lymphangioma.[2] Although open transabdominal adrenalectomy had been applied for the surgical removal of comparatively large adrenal cystic tumor, laparoscopic surgery is becoming standard for all benign adrenal tumors even in those with large size more than 8 cm because of its reduction in blood loss, lower requirements for analgesia, shorter hospital stay, and quicker recovery.[2]

In the present case, catecholamine titers of the cyst contents were extremely high although it was later revealed. Thus, there was a risk of exposure to catecholamines following cyst rupture. Costa Almeida et al.

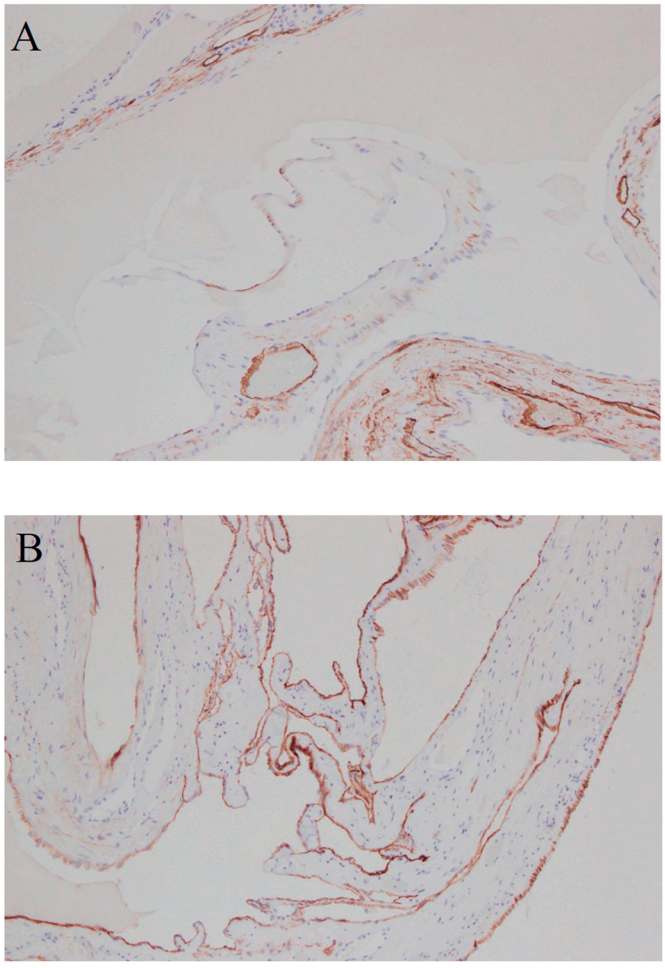

Fig. 2. Histopathology of the left adrenal cystic tumor shows multilocular cysts. Immunohistochemically, cyst covering cells show positivity for CD34 (A) and D2-40 (B). (hematoxylin and eosin staining, 100 ×).

Table 1
Catecholamine titers of the cyst contents.

	Present case	Reference value
Cortisol (μg/dL)	76.5	6.2–19.4
Renin activity (ng/mL/h)	0.5	0.2–2.7
Aldosterone (pg/mL)	4101	20–130
Adrenaline (pg/mL)	487623	<100
Noradrenaline (pg/mL)	131693	100–500
Dopamine (pg/mL)	1634	<30

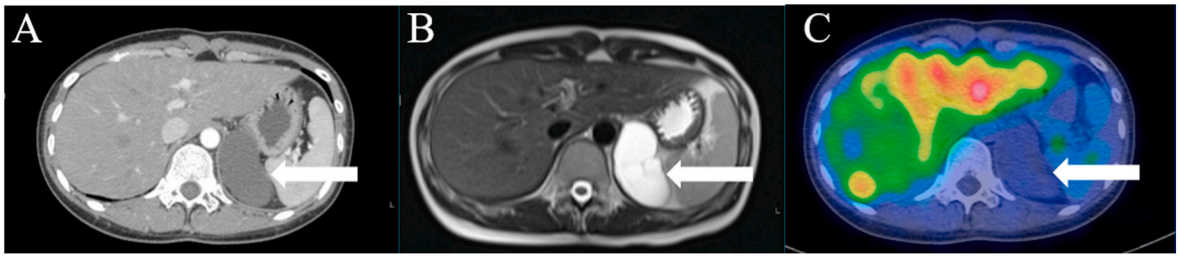

Fig. 1. (A) Contrast-enhanced abdominal computed tomography scan showed the presence of a left adrenal mass with a maximum diameter of 70 mm. (B) A plain T2-weighted abdominal magnetic resonance imaging; it indicated the presence of a left adrenal cystic tumor. (C) 131I-meta-iodobenzylguanidine scintigraphy showed no abnormal uptake in the left adrenal gland. (indicated by arrow).

reported intraoperative unintended rupture of a giant cystic pheochromocytoma which might lead to spillage of cancer cells.[5] Therefore, a surgical technique tailored to prevent cyst rupture was required in our present case as well.

Specifically, by storing and puncturing the large-sized tumor in an endoscopically inactive treatment tool, it was possible to both reduce the volume and remove the catecholamine-containing cyst without any exposure and possible to extract the gland without extending it.

Conclusion

As mentioned earlier, there is a possibility that adrenal lymphangioma which is difficult to differentiate from cystic pheochromocytoma preoperatively contain an extremely abnormal catecholamine titer. Due to its life-threatening consequences, unnecessary exposure to the extremely high catecholamine titers should be avoided. Therefore, our present approach described here might be reasonable for adrenal cystic tumor.

Author disclosure statement

No competing financial interests exist.

Acknowledgement

This research project received no specific grant from funding agencies in the public or commercial sectors.

References

1. Jacobi JD, Carballeira A, Fishman LM. Adrenal cysts: hormonal contents and functional evaluation. *Acta Endocrinol*. 1978;88:347–353.
2. Michalopoulos N, Laskou S, Karayannopoulou G, et al. Adrenal gland lymphangiomas. *Indian J Surg*. 2015;77:1334–1342.
3. Nigawara T, Sakihara S, Kageyama K, et al. Endothelial cyst of the adrenal gland associated with adrenocortical adenoma: preoperative images simulate carcinoma. *Intern Med*. 2009;48:235–240.
4. Andreoni C, Krebs RK, Bruna PC, et al. Cystic phaeochromocytoma is a distinctive subgroup with special clinical, imaging and histological features that might mislead the diagnosis. *BJU Int*. 2008;101:345–350.
5. Costa Almeida CE, Silva M, Carvalho L, et al. Adrenal giant cystic pheochromocytoma treated by posterior retroperitoneoscopic adrenalectomy. *Int J Surg Case Rep*. 2017;30:201–204.