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Oncology

A case of adrenocortical oncocytic neoplasm treated with laparoscopic adrenalectomy

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1. Introduction

Adrenocortical oncocytic neoplasms, i.e. adrenocortical oncocytomas, are very rare tumors that are usually nonfunctional and benign. Since the first description by Kakimoto et al. in 1986,[1] 162 cases have been reported.[2] A case of a left adrenocortical oncocytic neoplasm is described.

2. Case presentation

A 32-year-old woman who was followed for mammary adenoma every six months was found to have a left adrenal tumor on ultrasound examination in August 2016. She had no complaints and no abnormalities on physical examination.

Blood analyses, including biochemical test and endocrinological test, were within normal values. However, 24-h urine collection showed modest elevation of catecholamines except for adrenaline and metanephrine. CT and MRI showed a round left adrenal mass, with a diameter of 54 mm (Fig. 1). ^{123}I-adosterol scintigraphy showed no uptake in the tumor. We suspected the tumor of a paraganglioma with low endocrine activity or a malignant adrenal tumor based on these radiological examinations. Laparoscopic left adrenalectomy was performed with a transperitoneal approach. During the operation, the patient's blood pressures and heart rates were within normal ranges. Macroscopic examination showed that the color of the divided surface was mahogany. On microscopic examination, the tumor had a capsule and was well-circumscribed, however some of the tumor cells invaded to capsule. And most tumor cells were oncocytic cells containing eosinophilic granular cytoplasm (Fig. 2). On immunohistochemical analysis, the tumor cells were positive for SF-1, negative for 3β-hydroxysteroid, CYP11β1, and DHEAST. The cells were diffusely positive for mitochondrial stain. Therefore, the tumor was histologically diagnosed with adrenocortical oncocytic neoplasm.

The patient had an uneventful postoperative course and was discharged, and there was no recurrence 4 months after the operation.

3. Discussion

Adrenocortical oncocytic neoplasms are very rare tumors. Since the first description by Kakimoto et al. in 1986,[1] 162 cases have been reported.[2] These tumors are generally benign and they occur in all age (mean age at diagnosis 47 year-old, range 27–72 year-old).[3] They have been reported to occur more frequently in females (2.5:1) and in the left gland (3.5:1), are often detected incidentally by imaging during evaluations for unrelated problems.

Mearini et al. identified 147 cases of adrenal oncocytic neoplasms, and most were nonfunctional masses. In the present patient, blood analyses were within normal ranges, and she had no features of hormone excess, although a 24-h urine collection showed modest elevation of catecholamines except for adrenaline and metanephrine. Although these results did not fulfill the diagnostic criteria for pheochromocytoma and we performed the operation according as pheochromocytoma by way of caution, the patient's blood pressure and heart rate kept within normal ranges during the operation.

In the imaging of adrenocortical oncocytic neoplasms, it is important to differentiate the lesion from the adrenal adenoma. Most malignant lesions of adrenal gland show lipid-poor findings, whereas the majority of benign lesions indicate lipid-rich imaging in CT. And injected contrast material tends to wash out of benign adrenal lesions faster than malignant ones. However, the CT findings are generally nonspecific, and there is no characteristic imaging of adrenal oncocytic neoplasm. MRI also reveals no characteristic imaging.

An adrenocortical oncocytic neoplasm, histologically displaying

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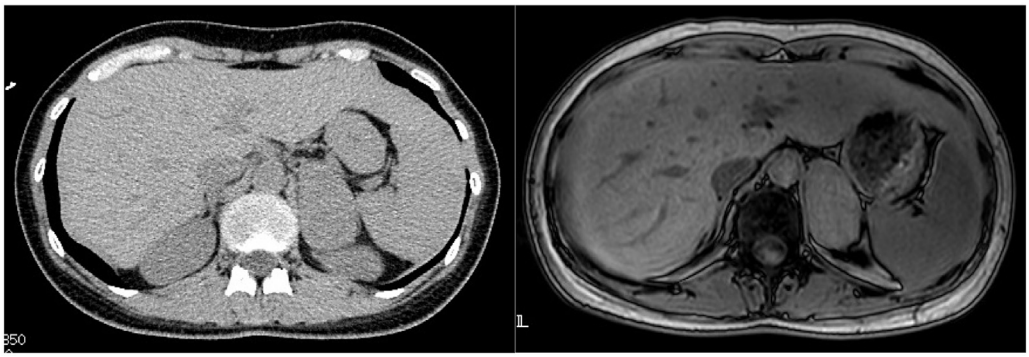

Fig. 1. CT and MRI of left adrenal mass. CT and MRI reveals a round left adrenal mass.

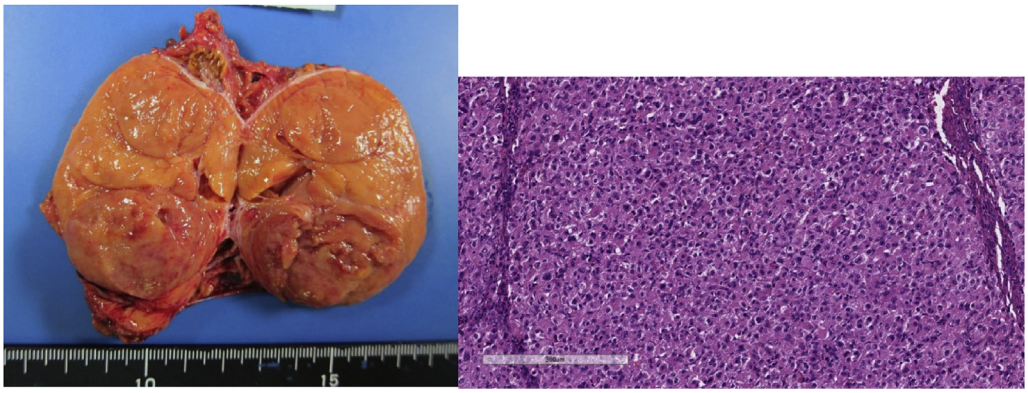

Fig. 2. Macroscopic surface and microscopic image of adrenocortical oncocytic neoplasm. The division surface of the left adrenocortical oncocytic neoplasm was mahogany (left). The tumor cells were oncocytic cells containing eosinophilic granular cytoplasm (right).

predominant cells with eosinophilic and granular cytoplasm, a high nuclear grade, and a diffuse architectural pattern, is diagnosed according to the Lin-Weiss-Bisceglia criteria.[4] According to this criterion, because of the tumor invasion for the capsule, the tumor of this patient was diagnosed as an uncertain malignant potential.

The surgical management of adrenocortical oncocytic neoplasms is adrenalectomy. Traditionally open adrenalectomy has been standard, but recent advances in endoscopic technique allow the laparoscopic adrenalectomy. In recent Robotic adrenalectomies for an adrenocortical oncocytic neoplasm are reported.[5] In spite of the advance of endoscopic surgical technique, the number of laparoscopic adrenalectomy for the adrenocortical oncocytic neoplasms is not large and there are only thirteen reports to the best of our knowledge (Table 1). Among these reports, the age of patients is relatively young (mean age is 34-year-old) and more than half its diameter of the tumor are 6cm or larger. Although a laparoscopic adrenalectomy is minimally invasive surgical procedure, the choice of laparoscopic approach to adrenocortical oncocytic neoplasm must be decided in a careful manner. Because most adrenocortical oncocytic neoplasms are detected at a large size and have a difficulty in preoperative diagnosis, we should take account of malignant tumors and assess the indication of laparoscopic approach. If a laparoscopic adrenalectomy is decided, it is important to convert to an open adrenalectomy when division of adhesion is difficult or the risk of capsular disruption is high. In addition, as with this case, it is necessary to prepare for pheochromocytoma if preoperative

Table 1
Cases of laparoscopic adrenalectomy for oncocytoma.

Author	Age	Sex	Side	Max diameter	Hormone production	Approach	Complication	Diagnosis	Ref.
Farkas A	34	male	right	6 cm	no	–	–	–	Orv Hetil 2005;146(27): 1453–58
Sharma N	47	female	left	9.5 cm	yes	transperitoneal	no	–	Indian J Pathol Microbiol 2008;51(4): 531–33
Eldahshan S	39	male	left	6 cm	no	transperitoneal		malignant	Arch Ital Urol Androl 2008;80(2): 82–84
Akatsu T	38	female	right	4 cm	yes	–	–	benign	J Endocrinol Invest 2008;31(1): 68–73
Kekis P	34	male	right	6 cm	no	transperitoneal	no	–	Int J Surg Case Rep 2012;3(7): 279–282
Qureshi AH	20	female	right	6 cm	no	–	–	–	J Coll Physicians Surg Pak 2014;24(12): 947–948
Sahin SB	23	female	left	2.7 cm	yes	–	–	benign	Case Rep Endocrinol 2014: 206890
Son SH	53	male	left	9.8 cm	yes	–	–	borderline	Ann Nucl Med 2014;28(1): 69–73
Yordanova G	9	female	–	–	–	–	–	benign	J Pediatr Endocrinol Metab 2015;28(5–6): 685–690
Lee HS	32	female	lert	1.8 cm	no	–	no	benign	Case Rep Surg 2016: 5790645
Kotoulova M	33	female	right	–	yes	–	–	–	Ceska Gynekol 2016;81(3): 228–232
Tartaglia N	44	female	right	3 cm	yes	transperitoneal	no	benign	Case Rep Surg 2016: 8964070
Current case	32	female	left	5.4 cm	no	transperitoneal	no	borderline	

catecholamine levels are high. If we perform laparoscopic adrenalectomy with these caution, according to our current report and past reports, laparoscopic adrenalectomy is safety, especially when the preoperative CT and MRI findings reveal a well-encapsulated tumor.

4. Conclusion

Recent advances in endoscopic technique allow the laparoscopic adrenalectomy. Most adrenocortical oncocytic neoplasms are detected at a large size and have a difficulty in preoperative diagnosis, therefore we should take account of malignant tumors and assess the indication of laparoscopic approach.

5. Consent

Appropriate informed consent has been obtained from the patient.

Conflicts of interest

None of the contributing authors have any conflict of interest, including specific financial interests or relationships and affiliations relevant to the subject matter or materials discussed in the manuscript.

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