A Functioning Adrenocortical Oncocytoma

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Adrenocortical oncocytes are exceptionally rare and most are benign and nonfunctioning. Only 3 cases of adenocortical oncocytes have been reported in the Korean literature and all of them were nonfunctioning. Herein, we report a case of a functioning adenocortical oncocyteoma in a 49-year-old man who presented with Cushing syndrome. (Korean J Urol 2009;50:401-403)

Key Words: Oxyphilic adenoma, Cushing syndrome

An oncocyoma is a benign tumor consisting of oncocytes in which the cytoplasm became eosinophilic due to the accumulation of abnormal mitochondria. Oncocytes develop in various organs and are frequently found in the salivary gland, the kidney, the thyroid gland, the parathyroid gland, and the hypophysis.1 However, adenocortical oncocytes have rarely been reported; just 25 cases and 3 cases have been reported in the foreign literature and domestic literature, respectively.2-4 Most of the oncocytes reported in the literature were nonfunctioning tumors and were incidentally found during a health examination.

In the present case, the authors found a left adrenal tumor in a 49-year-old male who visited the hospital as a result of Cushing syndrome and underwent an adrenalectomy. A histopathologic examination was performed after the operation, and as a result, the case was judged to be an oncocyoma. Here we report the case along with a bibliography.

CASE REPORT

A 49-year-old male visited the hospital because he had gained 2 kg over 4 months and had symptoms of truncal obesity and also facial edema that had progressed for 2 months.

![Enhanced abdominopelvic computed tomography. A 7.7 cm mass enclosed by a capsule was found on the left adrenal gland (A). Some necrotic tissue was found in the mass (B).](image-url)
There were no abnormalities on the results of a blood test, a serum electrolyte test, and a biochemical test. Twenty-four hour urinary free cortisol (UFC) was increased by 722.18 g (58-403 g) per day and did not decrease with the low-dose dexamethasone suppression test. Cushing syndrome was therefore diagnosed. An abdominal computed tomography (CT) scan was performed to locate the lesion and a 7.7 cm tumor accompanied by internal necrosis was observed on the left adrenal gland (Fig. 1). Positron emission tomography (PET) and CT were performed to ascertain whether the tumor was malignant and was metastasized to other organs. As a result, a hypertrophy was observed of the left adrenal lump but lesions suspicious of metastases were not found (Fig. 2). In the results of several tests performed to assess adrenal function, blood epinephrine, norepinephrine, vanillylmandelic acid, and total metanephrine were within normal levels.

We diagnosed the case as Cushing syndrome caused by the adrenal tumor and performed laparotomy and adrenalectomy because the malignant tumor could not be excluded clinically. The tumor did not seem to be metastasized to other organs at the time of the operation, and 7 days after undergoing the operation, the patient was discharged without intraoperative or

Fig. 2. Whole-body positron emission tomography/computed tomography (PET/CT) scan. A 7.7 cm hypermetabolic mass lesion with fluorodeoxyglucose (FDG) uptake was found in the left adrenal gland.

Fig. 3. The tumor was grossly well circumscribed with a dark brown color.

Fig. 4. Typical structure of an oncocytoma with abundant eosinophilic and granular cytoplasm. Nuclear atypia with enlarged nuclei was found (H&E, x200).

Fig. 5. Electron microscopy. Electron dense inclusion and closely packed mitochondria were found. Some destructed mitochondria with Golgi complex were found (x4,000).
postoperative complications. The extracted lump weighed 260 g and measured 10.0x7.5x4.7 cm. At the incisal surface, the tumor was yellowish and was surrounded by a well-defined capsule; on the inside, necrosis and hemorrhage were partially observed (Fig. 3). Light microscopy showed that the tumor was surrounded by a fibrous pseudo-capsule, and adrenocortical necrosis was seen in patches. Also, the tumor tissue consisted of polygonal cells, having abundant eosinophilic cytoplasm, in a regular form. Most nuclei were similar in size, but some were atypically large (Fig. 4). Electron microscopy showed that the oncotypic cytoplasm was filled with a great number of mitochondria; also, some lysosomes, Golgi bodies, small lipid particles, and glycogen particles were observed as is usual with oncocytes. Likewise, inclusion bodies of high electron density were observed in mitochondria (Fig. 5).

**DISCUSSION**

An oncocyto is a tumor consisting of polygonal cells that abundantly have eosinophilic granule cells, i.e., oncocytes. The oncocyte is defined as a cell in which abnormal mitochondria, which exclude other structures, accumulate. Mitochondrial accumulation and tumorigenesis have not been clarified yet, but there is a possibility that it is related to the inflammatory response, regression, or cellular aging. Many scientists infer that mitochondrial accumulation compensatorily causes functional loss. Oncocytes develop in various organs, but are especially frequent in epithelial cells of high metabolic activity. Namely, they mostly develop in the salivary gland, the kidney, the thyroid gland, the parathyroid gland, and the hypophysis.1

Adrenocortical oncocytes have rarely been reported; just 25 cases and 4 cases, inclusive of the present case, have been reported in the foreign literature and the domestic literature, respectively.2-4 With regard to functioning oncocytes, no cases have been reported in the nation. Even in other countries, just 5 cases have been reported:5-9 namely, one case where androgenic hormone was secreted in a female, one case where interleukin-6 was produced, and 3 cases of Cushing syndrome. The present case indicates Cushing syndrome accompanied by truncal obesity and facial edema.

In most cases, the adrenal tumor was found in process of the examination. The possibility of a metastatic tumor was ruled out because primary lesions were not observed on CT or PET, and the case was diagnosed as Cushing syndrome by assessing adrenal function.

The light microscopy and electron microscopy performed after the operation showed typical characteristics of oncocytes as reported elsewhere.5-10 The cellular structure of the oncocyto was compared with other adrenal tumors by using an electron microscope. In the case of the pheochromocytoma, a number of granules were bound to the cytomembrane of high electron density in neurosecretory granules and tumor cells. Benign adrenal adenomas are not filled with mitochondria. Such characteristics distinguish the oncocyto from others.

Nonfunctioning adrenal tumors, which are incidentally found, are open to dispute. On the other hand, surgical operations should be performed on functioning adrenal tumors. In addition, medical scientists should further study functioning oncocytes and be more aware of them, in consideration of their rareness.

**REFERENCES**