Adreno-Cortical Oncocytoma : A Case Report

Ibtissem Bouasker*, Ayoub Zoghlami*, Faten Farah Klibi**, Imen Smaali*, Mohamed Ali El Ouaer*, Rachida Zermani**, Chadli Dziri*.

* Service de Chirurgie Générale B, Hôpital Charles Nicolle, Tunis, Tunisie.
** Laboratoire d'Anatomie et de Cytologie Pathologiques, Hôpital Charles Nicolle, Tunis, Tunisie

I. Bouasker, A. Zoghlami, F. Farah Klibi, I. Smaali, M. A. El Ouaer, R. Zermani, C. Dziri.

L'adénome surrénalien à cellules oncocytaires : A propos d'un cas

LA TUNISIE MEDICALE - 2010 ; Vol 88 (n°05) : 353 - 356

I. Bouasker, A. Zoghlami, F. Farah Klibi, I. Smaali, M. A. El Ouaer, R. Zermani, C. Dziri.

Adreno-Cortical Oncocytoma: A Case Report

LA TUNISIE MEDICALE - 2010 ; Vol 88 (n°05) : 353 - 356

RÉSUMÉ

Prérequis : l'adénome surrénalien à cellules oncocytaires est une tumeur très rare, souvent bénigne et découverte fortuite, 46 cas sont rapportés dans la littérature médicale, elles sont souvent de découverte fortuite.
But: Rapporter un nouveau cas d'oncocytome surrénalien à potentiel malin.
Observation : Mr Y S âgé de 72 ans, sans rares tares. Il a présenté quelques jours avant son admission de douleurs de la fosse lombaire droite. L'échographie abdominale et la tomodensitométrie abdominale ont objectivé une volumineuse formation tissulaire de 13 cm envahissant le rein droit. Une résection en bloc emportant le rein droit et surrénal droite a été réalisée. L'examen anatomopathologique a conclu à un adénome surrénalien à cellules oncocytaires ave un potentiel malin. Les suites opératoires étaient simples. Il est réguliérement suivi à la consultation, il n'a pa réenté de récidive avec un recul de 8 mois.
Conclusion : l'adénome surrénalien à cellules oncocytaires est une tumeur trés rare. Seul l'examen anathomopathologique permet de psr le diagnostic positif. Le pronostic est souvent bon.

SUMMARY

Background : Adrenal oncocytoma is a very rare lesion, non functioning and benignin most cases. Only 46 cases have been reported in the medical literature.
Aim: This study aimed to report a new case of adrenal oncocytic tumor with uncertain malignant potential.
Case: A 72 year-old- man, consulted for renal fossa pain. Ultrasonography and omputed tomography scan revealed a large mass in the right adrenal gland with extension to the right kidney. A right adrenalectomy and nephrectomy was performed. The diagnosis of adrenal oncocytoma with malignant potential was confirmed by pathology. Patient had a well recovery and left hospital on the fifth day post operatively. He was followed up for 8 months, no tumor recurrence detected.
Conclusion: Adreno cortical oncocytoma is a rare tumor. The majority of reported cases had good prognosis.

Mots-clés
adénome surrénalien; tumeur

Key-words
Adrenal gland; oncocytoma; tumor

الغدّوم الحمضي في الكظرية : حالة واحدة

الباحثون : ابتسام بو عسكر - أيوب الزغلامي - فاتن فرح القليبي - إيمان السمعلي - محمد علي الواعر - رشيدة زرماني - شادلي الدزيري .

الهدف من هذه الدّراسة هو استعراض حالة غدّوم حمضي خبيث في الكظرية عند مريض عمره 72 سنة .أثبت التصوير بالصدى وجود كتلة كبيرة في الكلية اليمنى وقع استئصالها جراحيا و أثبت التشريح المرضي أنّها غدوم حمضي خبيث .نستنتج أن هذا الورم نادر لكّن إنذاره طيّب

Oncocytictumour of adrenal gland was defined as a neoplasm composed exclusively or predominantly of oncocytes which are large and polygonal cells with eosinophilic cytoplasm because of abnormal accumulation of mithochondria [1]. It was first described in 1986 by Kakimoto[2]. Only 46 cases have been reported in the literature [2- 6] most of them are non functional and benign [7]. We report a newcase of adrenal oncocytoma in 72-year-old man.

CASE REPORT

A 72-year-old man complained of right renal fossa pain since 6 days prior to admission. The patient denied any symptoms of palpitation, headaches, dizzinem or visual disturbance. No special signs were found by physical examination, blood pressure was 120/60 mmHg. Laboratory investigations revealed normal serum electrolytes. The urinary catecholamine levels were all normal. Hormonal studies were normal. Abdominal ultrasonography showed a large 13cmx10cmx8cm heterogeneous mass in the right adrenal gland with low level echo and with no calcifications. Abdominal computed tomography scan confirmed the right adrenal mass of 13cmx10cmx9cm, in the vicinity of the superior pole of the right kidney, which was with mixed density (figure 1 and 2).

Figure 1 : Abdominal computed tomography scan confirmed the right adrenal mass of 13cmx10cmx9cm, in the vicinity of the superior pole of the right kidney, which was with mixed density

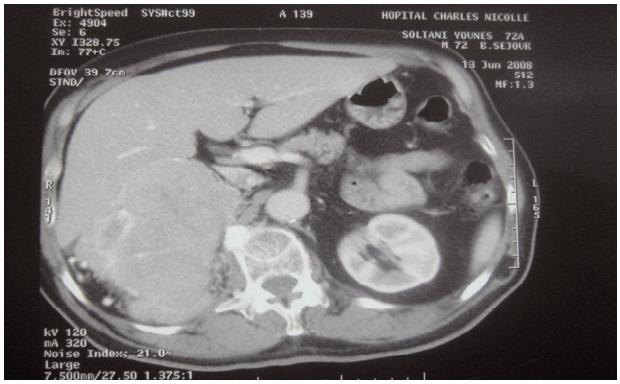
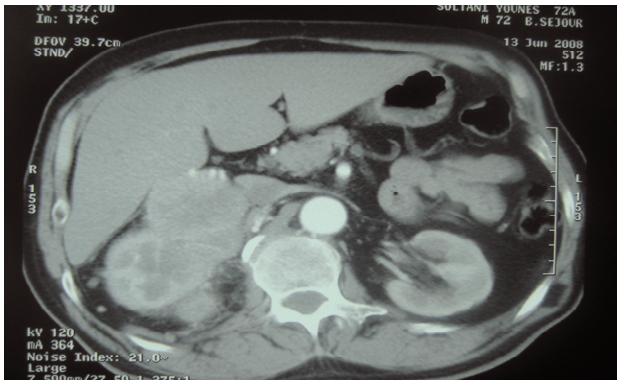


Figure 2 : Abdominal computed tomography scan confirmed the right adrenal mass of 13cmx10cmx9cm, in the vicinity of the superior pole of the right kidney, which was with mixed density



The postcontrast images in the arteriel phase show the presence of a rich vascularity (figure 3). At laparotomy, an adrenal mass was noted with extension to the right kidney. A right adrenalectomy and radical right nephrectomy was performed. When the mass was touched and removed blood pressure did not fluctuate. The tumor was brown, solid, well circumscribed, measured 16cmx9cmx11cm. Little focal haemorrhage was present. Microscopically (figure 4) the tumor was composed of polygonal oncocytes with abundant granular, eosinophilic cytoplasm. Cells were arranged in a diffuse pattern. No mitotic figures, neithero lymphovascular or capsular invasion was seen. The tumor was immunoreactive for cytokeratin (figure 5) and antimitochondrial antibodies, negative for PS 100 and chromogranin. The pathological diagnosis was adrenal oncocytoma without any malignant potential. The post operative course was uneventful and the patient was discharged on the fifth day. The patient was followed up for 8 months, no tumor or metastases were detected by CT scan.

Figure 3 : The postcontrast images in the arteriel phase show the presence of a rich vascularity

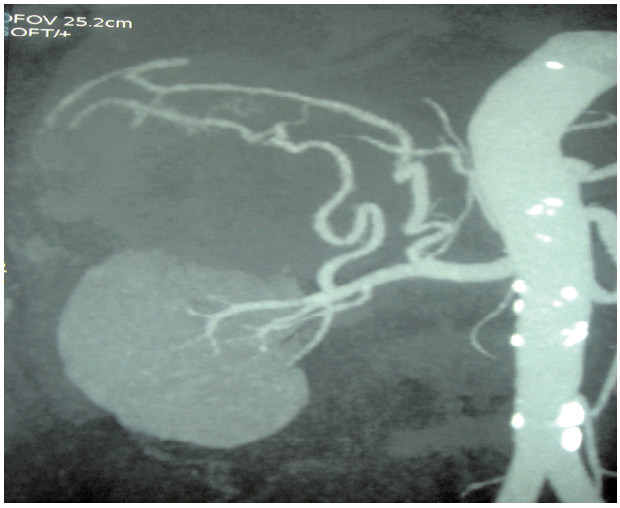


Figure 4 : Tumor cells have characteristic oncocytic aspects with abundant eosinophilic and granular cytoplasm with prominent nucleoli(hematoxylin and eosin, magnificationx200 and x400)

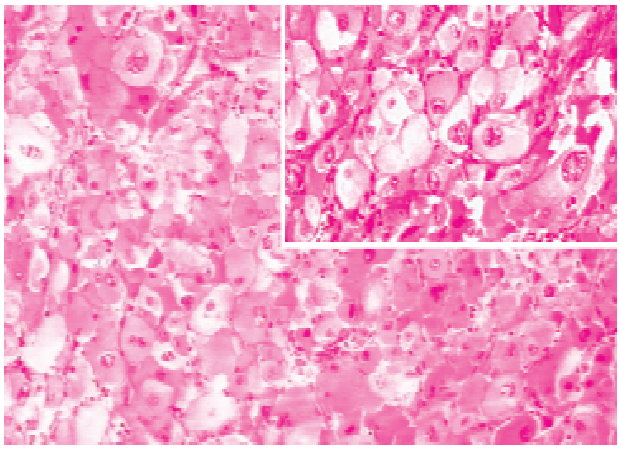
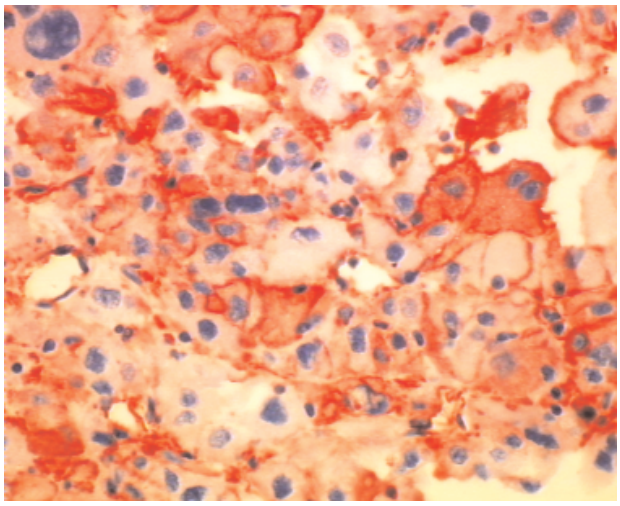


Figure 5 : The tumor cells cytoplasm showing focally positive immunoreactivity for Cytokeratin(magnificationx400)



DISCUSSION

We reported a non functioning oncocytoma of the adrenal gland, which was diagnosed on right renal fossa pain, in 72-year-old man. Abdominal computed scan revealed a right adrenal mass and removal was performed. Histopathological study confirmed the diagnosis of adrenal oncocytoma. Oncocytic neoplasms are composed of oncocytic cells which are characterized by having larges eosinophilic granular cytoplasm owing to the abberant accumulation of mithochondria [8]. Oncocytic tumour are extremely rare, only 46 cases have been reported and include: 24 oncocytomas, 6 oncocytic neoplasms of uncertain malignant potential and 16 oncocytic carcinoma [9]. The most commonly reported sites for oncocytic neoplasms are the thyroid, kidney and salivary glands [6, 10], found predominantly in adults [11, 12]. Only two cases was reported in child [12, 13], female are more concerned than male patients with a female ratio of 7/4 [1]. Most cases have been benign, non functioning and incidentally detected usually by imaging examination in regular health examination or during investigation for abdominal pain or unrelated symptoms [12, 14, 15]. Gargah et al reported a 12-year-old girl with pseudo precocious puberty induced by adrenocortical oncocytoma. Kabayegit et al reported a 31-year-old man presented with Cushing syndrome. The same case was reported by Xiao et al. Our patient had a non-functioning adrenocortical oncocytic neoplasm. Computed tomography and magnetic resonance images have no special appearance [14, 16]. In 2008, Kim DJ et al [17] has reported for the first time on the F-fluorodeoxyglucose positron emission tomography finding in a case of adrenocortical oncocytoma, and the cause may be the intense hypermetabolism of glucose and the numerous intracellular mitochondria. The pre operative diagnosis is often non fuctioning tumor of adrenal gland. An adrenalectomy can be often performed. With the development of laparoscopic technique, adrenal tumour can be resected under laparoscopic procedure [14, 18]. However, there is still contreversely regarding the appropriateness of laparoscopy for the adrenal tumors resection in cases of uncertain malignant potential [14]. Our case was a large tumor 13cmx10cmx9cm, removed by laparotomy. The diagnosis was established by histological and immunhistochemical studies. Most of the adrenal oncocytoma were well circumscribed, capsulated, ranging from 2 to 20 cm [14, 15, 19]. In our case the oncocytoma was of 16cmX11cmx9cm. The tumor cells are highly eosinophilic and granular, which attributed to the presence of numerous mithochondria. Immunocytochemical study revealed usually strongly positive for antimitochondrial antibodies, and frequently, vimentin and keratin is identified [11, 12, 20]. Generally, oncocytoma is considered as a benign tumor, but a malignant variant with local invasion and distant metastases was described in 1991[21]. Four cases of adrenal oncocytic carcinoma were described in 2002[11]. It is important to discriminate adrenocotical adenoma and carcinoma. In 1989 weiss et al proposed histological criteria which was modified by Bisceglia et al in 2004[22]. According to this criteria: the presence of one major criteria (high mitotic activity, atypical mitoses or venous invasion) indicating malignancy, one to four minor criteria (Large size, necrosis, capsular or sinusoidal invasion) indicating uncertain malignant potential (borderline) and the absence of criteria indicates a benign tumor. Others markers such as Ki-67(+) has been reported to be helpful in distinguishing adrenocortical adenomas from carcinomas [23]. In our case, the tumor have a large size, therefore this case was diagnosed as oncocytic neoplasm with uncertain malignant potential.

CONCLUSION

We present a case of 72-year-old man with non-functioning adrenocortical oncocytoma, greater than 6 cm. Adrenalectomy and nephrectomy were performed. It was a benign tumor with potential malignant feature. Our patient was good after surgery and symptoms free, 8 months post operatively.

Références

1. Xiao GQ, Pertsemlidis DS, Unger PD. Functioning adrenocortical oncocytoma. A case report and review of the literature. Ann Diag Pathol 2005; 9: 295-97
2. Kakimoto S, Yushita Y, Sanefugi T, Kondo A, Fujishima N, Kishikawa M, et al. Non-hormonal adrenocortical ademona with oncocytoma like appearances. Acta Urol Jpn 1986; 32: 757-63
3. Logasundaram R, Parkinson C, Donaldson P, Coode PE. Co-secretion of testosterone and cortisol by a functional adrenocortical oncocytoma. Histopathology 2007; 51: 418-20
4. Waters PR, Haselhuhn GD, Gunning WT, Phillips ER, Selman SH. Adrenocortical oncocytoma: two case reports and review of literature. Urology 1997; 49: 624-628.

5. Sasano H, Suzuki T, Sano T, Kameva T, Sasano N, Nagura H. Adrenocortical oncocytoma: a true non-functioning adrenocortical tumor. Am J Surg Pathol 1991; 15: 949-956
6. Kitching PA, Patel V, Harach HR. Adrenocortical oncocytoma. J Clin Pathol 1999; 52: 151-153.
7. Sundarama S, KuruvillaS, Varghese RG et al. Adrenocortical oncocytoma : a case report. Indian J Pathol Microbiol 2004; 47:387-9
8. Chang A, Harawi SJ. Oncocytes, oncocytosis, and oncocytic tumors. Pathol annu 1992;27:263-304
9. Kabayegit OY, Soysal D, Oruk G et al . adrenocortical oncocytic neoplasm presenting with cushing's syndrome: a case report. J Med Case Reports. 2008; 2:228
10. Lee R, Al-Ahmadie H, Boorjian SA et al. A case of incidental adrenocortical oncocytoma. Nat Clin Pract Urol 2006;3:618-21
11. Hoang MP, Ayala AG, Albores- Saavedra J. Oncocytic adrenocortical carcinoma: A morphologic immunohistochemical and ultrastructural study of four cases. Mod Pathol 2002;15:973-78
12. Gargah T, Kaabar N, Sayed S et al. Adrenocortical oncocytoma: a case report and review of literature. J Ped Surg 2008; 43:E1-E3.
13. Guym-Pause F, Bongiovanni M, Wildhber B et al. Adrenocortical oncocytoma in a child.Pediatr Blood Cancer. 2008;50:718-21
14. Tian XJ, Huang Y, Lu J et al. Laparoscopic adrenocortical oncocytoma resection. Chin Med J 2008 ; 121 : 862-64.
15. Germizadeh B, Norouzzadeh B, Bolandparvaz et al. Functionning adrenocortical oncocytoma : A case report and review of litterature. Indian J Pathol Microbiol 2008;51:237-9
16. Gandaras EJ, Schwartz LH, Panicek DM et al. Case report. Adrenocortical oncocytoma/ CT and MRI findings. J Comput Assist Tomogr 1996;20:407-709
17. Kim DJ, Chung JJ, Ryu YH et al. Adrenocortical oncocytoma displaying intense activity on F-FDG-PET: A case report and a literature review. Ann Nucl Med 2008;22:821-24
18. Modi P, Goel R, Kadam G. Retroperitoneoscopic partial adrenalectomy for large adrenocortical oncocytoma. J Endourol 2007;21:419-422
19. Botsios D, Blouhos K, Vasilias K et al. Adrenocortical oncocytoma : a rare tumor of undefined malignant potential : report of a case. Surg Today 2007;37:612-7
20. Poretti D, Mazzarol G, Bonomo G et al. Adrenocortical oncocytoma: case report. J Clin Imag 2003;27:426-30
21. El-Naggar AK, Evans DB, Mackay B. Oncocytic adrenal cortical carcinoma. Ultrastruct Pathol 1991;15:549-556
22. Bisceglia M, Ludovico O, Di Mattia A et al. adrenal oncocytic tumors: report of 10 cases and review of the literature. Int J Surg Pathol 2004; 12: 231-43
23. Stojadinovic A, Brennan MF, Hoos A et al. Adrenocortical adenoma and carcinoma : histopathological and molecular comparative analysis. Mod Pathol 2003;16:742-51