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Inflammatory myofibroblastic tumor of the adrenal gland: A case report

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ABSTRACT

Inflammatory Myofibroblastic Tumor (IMT) occurring in the adrenal gland is extremely rare, and pathologic examination is the gold standard for confirming the diagnosis. We report a case of IMT of adrenal origin in a patient whose diagnosis was confirmed by pathological examination after surgical resection of the tumor. Although previous studies have reported an overall favorable prognosis for IMT, regular and long-term follow-up is necessary.

1. Introduction

Inflammatory Myofibroblastic Tumor (IMT) is a rare mesenchymal tumor, with a higher prevalence in the lungs. Its occurrence in the urinary system, particularly the bladder, is relatively common, whereas IMT in the retroperitoneum is exceedingly rare.[1,2,3] There is a lack of available reports on adrenal IMT. This case report presents an instance of adrenal IMT.

2. Case presentation

The patient, a 38-year-old man, was diagnosed with a right adrenal mass during a physical examination four months ago. Upon admission to the outpatient clinic, no positive signs were observed during the physical examination. A subsequent CT scan revealed a mass in the right adrenal region measuring approximately 5.7cm × 7.8cm, exhibiting marginal enhancement (Fig. 1A and B). Laboratory tests showed no abnormalities. The patient underwent laparoscopic right adrenalectomy, and the postoperative specimen exhibited a significant tissue mass in the right adrenal gland measuring 10cm × 7cm × 4cm with grayish-yellowish appearance and no hemorrhage or necrosis(Fig. 1C). Pathological examination confirmed the presence of an inflammatory myofibroblastic tumor (IMT) in the right adrenal gland(Fig. 1D and E). Immunohistochemical analysis showed a specific pattern of antigen expression (Fig. 1F): CD117 (−), CD34 (−), SMA (−), Caldesmon (−), Calponin (−), Ki-67 (10 %), P53 (−), CD99 (−), BCL-2 (−), S100 (−), VIM (+), CK (−), SDHB (+), ALK (+). CD30 (−), CD138 (−), CD38 (scattered +), EMA (plasma cell +), β-catenin (−), Myo-D1 (−), Desmin (+), LCA (+). The patient's 2-year postoperative follow-up showed no tumor recurrence.

3. Discussion

In related studies, Liu et al.[4] found a 5-year overall survival rate of 77 % in adult IMT patients, with 42.1 % experiencing recurrence or metastasis after radical surgical resection.[5] Chen et al. reported a 43.5 % positivity rate of ALK in the genitourinary system, consistent with the positive ALK expression in the reported patient.[6] Previous articles have suggested that ALK-positive expression is associated with recurrence and metastasis of IMT, with 45 % of ALK-positive patients recurring, compared to 20 % of ALK-negative patients.[7] The pathogenesis of IMT remains unclear, possibly involving factors such as EBV infection and autoimmune diseases.[8] IMT's clinical manifestations are nonspecific,[9] with cases often incidentally detected during health check-ups. Surgical resection currently stands as the primary treatment for IMT, and postoperative pathological diagnosis is crucial for confirming the disease. Notably, thorough microscopic examination reveals characteristic features of IMT, including proliferating spindle-shaped fibroblasts, accompanied by inflammatory cell infiltration and germinal center formation.[10] In China, surgical resection remains the mainstay treatment for IMT. However, research on ALK inhibitors is gaining traction as a potential new approach, with prospective phase 2 clinical trials (EORTC90101) demonstrating promising outcomes.[11]

4. Conclusions

In conclusion, adrenal IMT cases are rare, and the diagnosis can be

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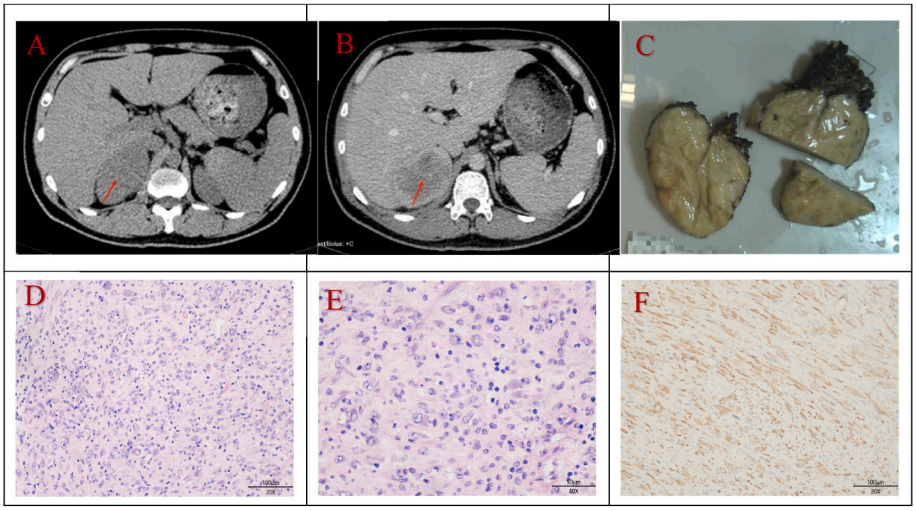

Fig. 1. Imaging images, pathology findings, immunohistochemical staining. A: CT plain scan imaging; B: CT contrast-enhanced scan imaging, exhibiting marginal enhancement. C: the photographs of the right adrenal gland specimen, measuring 10cm × 7cm × 4cm with grayish-yellowish appearance and no hemorrhage or necrosis. D: pathology results (low magnification). E: pathology results (high magnification). F: ALK immunohistochemical staining.

significantly enhanced through ALK gene rearrangement testing. Positive ALK expression may impact patient prognosis, although overall outcomes for IMT patients are favorable. Ongoing long-term follow-up remains essential post-surgery.

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Declaration of competing interest

All authors declare no conflict of interest.

References

1. Dong J, Ji ZG, Xu WF, et al. Report of four cases of adult renal inflammatory myofibroblastoma with literature review. *Int J Surg*. 2016;43(9):622–625.
2. Ariafar A, Ahmed F, Khorshidi A, et al. Inflammatory myofibroblastic tumor of the right kidney mimicking a locally advanced renal carcinoma: a case report. *Journal of Kidney Cancer and VHL*. 2022;9(4):14–18.
3. Harik LR, Merino C, Coindre JM, et al. Pseudosarcomatous myofibroblastic proliferations of the bladder: a clinicopathologic study of 42 cases. *Am J Surg Pathol*. 2006;30(7):787–794.
4. Liu X, Gong C, Zhang J, et al. Clinicopathological analysis and treatment of adult patients with inflammatory myofibroblastic tumor: a 15-year single-center study. *Cancer Research and Treatment*. 2023;55(3):1001–1010.
5. Alaggio R, Cecchetto G, Bisogno G, et al. Inflammatory myofibroblastic tumors in childhood: a report from the Italian Cooperative Group studies. *Cancer*. 2010;116 (1):216–226.
6. Chen B, Xu H, Guo YC, et al. Clinicopathology and prognosis of inflammatory myofibroblastoma of the genitourinary system. *J Mod Urol*. 2021;26(2):112–115+ 146.
7. Coffin CM, Watterson J, Priest JR, et al. Extrapulmonary inflammatory myofibroblastic tumor (inflammatory pseudotumor). A clinicopathologic and immunohistochemical study of 84 cases. *Am J Surg Pathol*. 1995;19(8):859–872.
8. Coffin CM, Humphrey PA, Dehner LP. Extrapulmonary inflammatory myofibroblastic tumor: a clinical and pathological survey. *Semin Diagn Pathol*. 1998; 15(2):85–101.
9. Wang Y, Qi X, Xue P, et al. Inflammatory myofibroblastic tumors in the kidney and abdominal wall mimicking malignancy: a case report. *Medicine (Baltim)*. 2018;97 (34), e11994.
10. Liu X, Zhang XW, Wang HJ, et al. Clinical characterization of 11 cases of inflammatory myofibroblastoma in adults. *Chin J Cancer*. 2018;28(6):424–428.
11. Schöffski P, Kubickova M, Wozniak A, et al. Long-term efficacy update of crizotinib in patients with advanced, inoperable inflammatory myofibroblastic tumour from EORTC trial 90101 CREATE. *Eur J Cancer*. 2021;156:12–23.