Metastatic Malignant Struma Ovarii

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Struma ovarii is a monodermal ovarian teratoma, composed mainly of differentiated thyroid tissue. It is a rare condition, representing less than 1% of all ovarian tumors. Almost always benign, malignant features present in only 5–10% of cases. We report the case of an adolescent with metastatic malignant struma ovarii with aggressive biologic behavior.

PATIENT DESCRIPTION

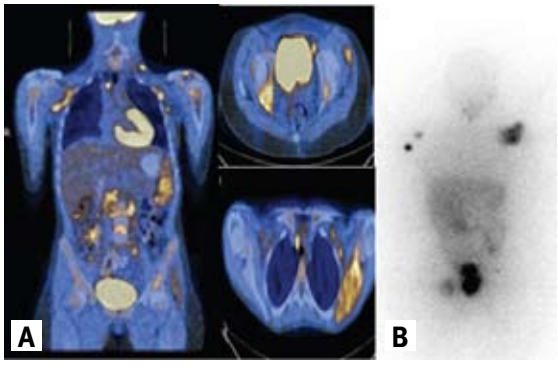
A 15 year old girl was admitted with the complaint of pain in the lower abdomen and pelvic discomfort. Abdominal ultrasonography showed a huge mass arising from the right ovary. A right oophorectomy was performed and macroscopic pathological examination revealed a tumor measuring 7 x 11.5 x 16.5 cm. Microscopic examination confirmed struma ovarii, a monodermal teratoma with positive thyroglobulin staining but without lymphovascular or capsular invasion.

Three months later, the patient complained of pain in the right hip and a lump in her posterior scalp. Brain computed tomography demonstrated a subcutaneous mass with destruction of the occipital bone and the appearance of intracranial expansion. The mass was excised and pathological examination confirmed follicular carcinoma of the thyroid with positive staining for thyroid transcription factor-1 and thyroglobulin. Neck ultrasonography was normal. The patient was clinically euthyroid.

Positron emission tomography-fludeoxyglucose F 18 scan showed diffuse pathological uptake in the bones (right acetabulum, left clavicula, left scapula), spleen, lymph nodes (cervical, supraclavicular, mediastinal, axillary, retroperitoneal, pelvic and inguinal), and subcutaneous nodules in the left occipital region and right neck [Figure A].

Total thyroidectomy was performed and the pathological report showed normal thyroid tissue. During the 4 weeks without levothyroxine after the surgery, while waiting for thyroid-stimulating hormone levels to rise, the patient received palliative radiotherapy of 40 Gy to the right acetabulum and the left scapula, with marked clinical improvement. The postoperative I-131 scan showed 1% uptake after 2 and 24 hours, followed by I-131 treatment with 150 mCi. The post-treatment I-131 whole-body scan showed pathological uptake in bone metastases, paratracheal lymph nodes, upper neck and left parotid [Figure B]. Our plan was to continue treatment with I-131 and consider biological treatment in the near future.

[A] PET-FDG and **[B]** post-treatment whole-body iodine scan highlight a pathological uptake in the metastases.



COMMENT

The first description of follicular thyroid tissue in the ovaries was made in 1889 by Boettlin. A differential diagnosis of malignant struma ovarii from a benign ovarian teratoma is difficult to make. It requires lymphovascular tumor invasion, recurrence or metastases, or the typical cytopathological features of papillary thyroid cancer. The follicular variant of papillary thyroid carcinoma in struma ovarii exhibits the same morphological and immunohistochemical profile as the follicular variant in the thyroid.

Our case is different from those reported in the literature for several reasons: Firstly, struma ovarii usually presents during the reproductive years, in the fifth decade on average [1]. It rarely appears before puberty. Only a few cases of struma ovarii in patients between age 14 and 16 have been described [2,3]. Secondly, metastatic spread by local invasion, peritoneal, lymphatic pathways, and hematogenic dissemination to brain and lungs have been reported, but only 11 cases of bone metastases from malignant stuma ovarii have been described in the

literature [4]. Thirdly, several studies in the literature have reported high sensitivity (up to 98%) and specificity (up to 95%) of PET-FDG in metastatic differentiated thyroid carcinoma [5]. Little is known about the value of PET-FDG in malignant struma ovarii. In our case, PET/CT was performed and showed diffuse pathological uptake, which indicates aggressive disease. PET/CT showed additional uptakes that were not apparent on the post-treatment I-131 scan, leading us to think of different approaches to the disease, not relying solely on radio-iodine treatment but including additional treatment such as biological therapy.

PET-FDG = positron emission tomography-fludeoxyglucose F 18

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