Primary adenocarcinoma of the seminal vesicle

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Abstract

Primary adenocarcinoma of the seminal vesicle is a rare condition with only about 60 cases described in the literature. The unusual characteristics of this disease makes diagnosis difficult and treatment strategies differ as there are no specific guidelines available. This report presents a case of adenocarcinoma of the seminal vesicle with lung metastases in which surgical and chemotherapeutic treatments have been carried out. The MVAC dose dense regimen following local resection seems effective in this scenario and may be used in the treatment of this disease.

Case Report

A 43-year-old man presented with azoospermia, perineal insensitivity, lumbar pain and rectal pus he had been suffering from for 6 months. Clinical examination revealed small testicles and the digital rectal examination showed an enlarged prostate with no fixed adenoma. Prostate specific antigen (PSA) was 1.52 ng/mL.

Prostatic MRI demonstrated non-specific abnormalities with: an enlarged prostatic gland, heterogeneous signal in left central and transitional parts of the prostate and loss of seminal-prostatic and deferent differentiation.

Transrectal biopsies of the prostate revealed a poorly differentiated carcinoma, probably of glandular origin. The immunohistochemical results are presented in Table 1. Based on these data, the diagnosis of a poorly differentiated carcinoma of probably urothelial origin with massive prostate extension has been established.

Complementary evaluation with thoracic CT revealed a nodule in the left superior lobe of the lung. PET scanner did not show any other metastasis. At that point, the patient experienced an exacerbation of the symptoms with hematuria and perineal pain. Due to fast clinical evolution and risk of incomplete resection of the lung nodule, the patient underwent primary chemotherapy. He received six cycles of MVAC dose dense regimen (Methotrexate 30 mg/m^2 D1, Cisplatin 80 mg/m^2 D2; Vinblastine 3 mg/m^2 D2, Doxorubicin 30 mg/m^2 D2) once every 15 days.

The patient had an excellent clinical response after the first chemotherapy cycle. An imaging response was obtained after the fourth cycle with partial response at the pelvis and complete pulmonary response.

Dose reductions of Cisplatin at 50 mg/m^2 were made during the fifth and sixth cycles because of moderate renal insufficiency (creatinin: 166 umol/L *vs* 85 umol/L at Cycle 1).

One month after the end of chemotherapy, the patient underwent surgical resection, viz., a cystoprostatectomy with an extended pelvic lymphadenectomy and an enterocystoplasty.

Final histopathological assessment showed a carcinoma proliferation centered on the seminal vesicles, negative for CK7, CK20, PSA, P63 and P504S. The lesion was classified as an adenocarcinoma of the seminal vesicles with invasion of the prostate and the lower part of the bladder. The margins of the resected specimen were described as negative, without lymph nodes metastases.

Two months after the surgery, the patient was still under observation when a new, 10-mm lung nodule was found, which grew to 21 mm in two months afterwards. He underwent atypical complete resection of the nodule in the left superior lobe and the immunohistochemistry confirmed a metastasis of an adenocarcinoma of the seminal vesicle.

The patient subsequently regained control over the disease and at the last follow up visit, he was in persistent complete remission 4 years after the pulmonary metastasectomy.

Discussion

Primary tumors of the seminal vesicles are a rare and poorly understood malignancy.[1] Symptoms are non-specific and difficult to differentiate from other retrovesical space tumors. The first histologically confirmed case of carcinoma of the seminal vesicle dates back to 1925 in Lyon, France and was reported by Lazarus in 1946.[2] The incidence cannot be known because of the small number of cases. Only about 60 cases have been reported in the literature so far.[3] Our review of the literature published since 2000 describes 12 cases, of which only 3 are metastatic.

At the time of diagnosis, age range goes from 19 to 90 but we could not find an accurate age average in the literature. In most cases, the disease frequently appears after 50 years of age.[3,4]

On the other hand, risk factors have not been formally described. In some cases, renal agenesis isconcomitant, even though the reason of this association remains undetermined.

Diagnostic difficulties include the absence of symptoms at an early stage and the lack of well-defined diagnostic criteria.[5,6] Symptoms in most cases are not specific, commonly: hematuria, hematospermia, pelvic/perineal pain and dysuria.

Digital rectal examination is an important part of the physical examination, especially to rule out tumors that may be located in the prostate.

CT scan, MRI and transrectal ultrasonography are useful but there is no standard protocol. In our case, the diagnosis was not obvious from the images (prostatic MRI and transrectal image for guided biopsy).

Immunostains and histomorphology are helpful to confirm the diagnosis. Immunohistochemistry is the key and

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Table 1. Immunohistochemical analysis of prostate biopsies in patient.

Laboratory parameter	Result	Conclusion
KL1, CK 903, P504S, CD99	Positive	
Cytokeratin subsets 7 (CK7)	Negative	
Epithelial membrane antigen (EMA)	Negative	
PSA, PSAm, P501S	Negative	Reject prostatic origin
CD30, OCT3/4	Negative	Reject germinal tumor
Desmin	Negative	Reject rhabdomyosarcoma
CD57, synaptophysin, chromogranin	Negative	Reject neuroendocrine carcinoma

shows that these tumors are PSA and PSAP negatives. Cytokeratin profile is usually CK7 positive and CK20 negative. In the majority of cases, CA125 is positive but was not performed in our case. CA125 can distinguish seminal vesicle adenocarcinoma from other tumors such as prostatic, bladder and rectal adenocarcinomas, because of its negativity.[7]

Due to the rarity of this disease, multidisciplinarity is recommended, while treatment guidelines are not available for the same reason.

Surgical excision is the mainstay treatment. Approach ranges from local to radical excision depending on the disease extension. Pelvic lymphadenectomy is also recommended.[1]

Various adjuvant strategies are described. In addition, radiotherapy can be used in case of positive margins or incomplete resection.[5,8] Hormonal therapy has been used either in adjuvant or palliative treatment but this indication must be assessed.[9] Also, chemotherapy treatment has been used with only 6 cases reported.[1,4,5,9-11]

In two cases, patients received adjuvant chemotherapy and in 4 cases, patients received palliative chemotherapy in metastatic indication. Various regimens were used with one drug or multiagent regimens: Folfox (Folinic acid-Fluorouracil-oxaliplatin), Docetaxel, Cisplatin-gemcitabine.

Our presented case is the only one in which the patient has received MVAC dose dense regimen and underwent surgery after the chemotherapy treatment.

Our patient has demonstrated a noticeable response to the 6 cycles of MVAC dose dense regimen, with a complete pulmonary response and partial pelvic response. Progression appeared in the second month after the local surgical treatment and it was the first lung metastasectomy ever described.

Time of progression is variable in the literature, with ranges from 2 to 21 months. Our patient exceeds all expectations with a disease-free survival of 48 months.

Long term prognosis for this disease is reported as extremely poor. However, in many cases, survival rates have not been released or patients had died before progression.[3] In our case, the patient has a much better prognosis than described in the literature, probably due to a slow growing tumor.

Conclusions

Our report shows the complexity of the diagnosis, due to the lack of specific signs and symptoms. Final diagnosis was made retrospectively with the histopathological assessment of the resected piece. Efforts to establish biomolecular patterns have yet to be carried on, in order to clarify diagnosis and therefore adapted treatment.

Furthermore, this is the first case of metastatic adenocarcinoma of the seminal vesicle in which chemotherapy had been used before the surgical treatment. In this way, MVAC dose dense regimen appears to be an interesting option in the treatment of this disease.

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