

LIVER RESECTION FOR METASTASIS OF ADENOID CYSTIC CARCINOMA OF THE LACRYMAL GLAND: A CASE REPORT AND LITERATURE REVIEW

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ABSTRACT

Adenoid cystic carcinoma (ACC) is a rare malignant tumor. The first description of the ACC dates back to 1856 by Billroth. The ACC of the lacrimal glands are very rare. They are known for their locally aggressive character. Liver metastases are almost exceptional. We report the case of a patient with hepatic metastasis ACC. After eye enucleation of the primitive lacrimal gland ACC, the patient received external adjuvant radiotherapy. Three years later, she presented an upper abdominal pain. CT scan and MRI showed a liver tumor and the diagnosis of intrahepatic cholangiocarcinoma was made regarding imaging features. A left hepatectomy was performed; histology concluded to a liver metastasis of ACC. The immediate post-operative course was uneventful and no adjuvant treatment was decided. Thirty four months later, the patient presented with lung metastasis. Liver metastasis of ACC are very rare. Extended liver surgery is recommended especially for isolated liver metastasis with a long free gap between primary tumor and the liver metastasis.

Keywords: Adenoid cystic carcinoma; Hepatic metastasis; Lacrymal gland.

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INTRODUCTION

The adenoid cystic carcinoma (ACC) represents less than 1% of tumors of the head and neck (1) and 1 to 2% of orbital tumors (2). The first description of the ACC dates back to 1856 by Billroth (3). It affects mainly the salivary glands and less frequently the lacrimal glands (4). The ACC of the lacrimal glands is characterized by a local malignancy. The infiltration of nervous nets is often found. The metastasis are rare, and when they exist, they include by order of frequency: Lungs, brain and bones. The hepatic metastases are rarely described, and exceptionally isolated. The resection of hepatic metastasis remains controversial (5). We report the case of a patient, carrier of a hepatic metastasis of a lacrymal ACC operated 04 years before.

OBSERVATION

In January 2011, a 63 years old female patient was diagnosed with a tumor of the right lacrymal gland tumor treated by eye enucleation. Histological study concluded to an ACC. The patient received an external 60 grays radiotherapy beam in post-operative.

Four years later, she performed an abdominal ultrasound, which revealed the presence of a heterogeneous liver mass within the left liver. The CT- scanner found an hypoattenuating mass on unenhanced CT (**Figure 1**), less enhancement than the surrounding liver on the arterial phase and become hyperattenuating than the parenchyma at the portal venous phase. Liver MRI was performed, found a huge polylobed left liver mass of 96/80 mm with high T2 signal intensity, centered by the bile

ducts dilated, diffusion restriction on DWI, low T1 signal intensity with the same enhancement as on the CT scan. The diagnosis of intra hepatic cholangiocarcinoma has been strongly suspected.



Figure 1: CT scan: CT arterial time: hypodense mass slightly enhanced, infiltrating appearance modifying the contours of the liver

A hepatic resection was indicated. A left hepatectomy was performed (**figure 2**). An extra glissonian control of the left pedicle was practiced and the liver resection was conducted without clamping.



Figure 2: surgical specimen, left hemi-hepatectomy

The histological study found a neoplastic proliferation of a poorly limited epithelial nature made of hollow cavities clusters, of tubular structures and cords. The stroma was fibrous. Cytonuclear atypia were present as well as mitosis.

Immunohistochemistry showed a positivity of the tumor cells for the cytokeratin 7, the CD117 and the EMA were found. The Ki67 was 60%. The final diagnosis was a hepatic metastasis of an ACC of the lacrymal gland. (**Figures 3, 4**)

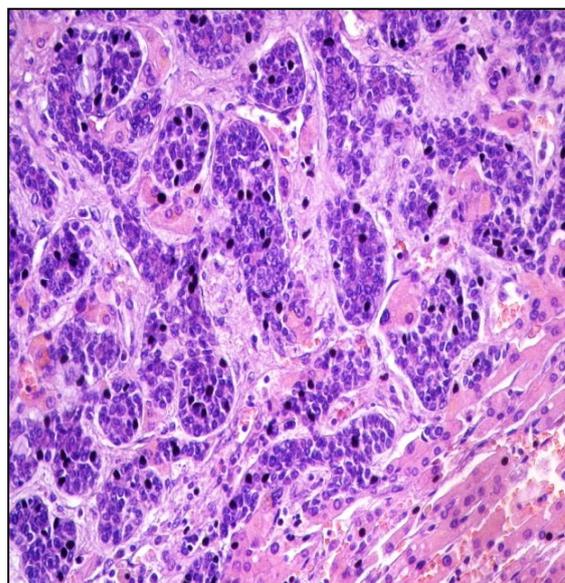


Figure 3: (HE x 200) : liver metastasis of ACC

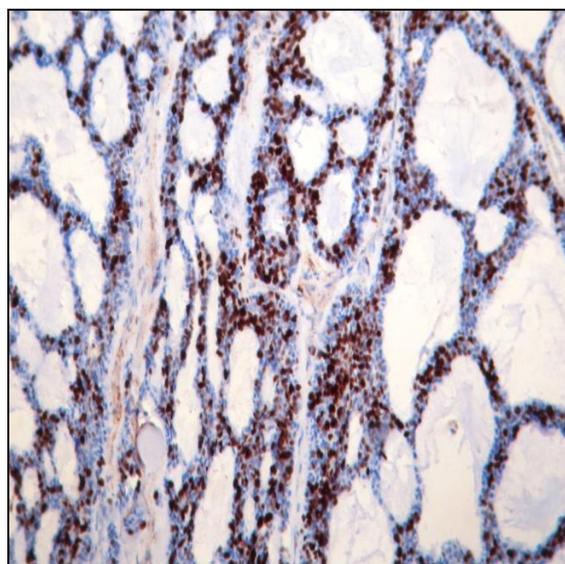


Figure 4: (IHC x 200): Ki67 tumor cells' nuclear staining (> 60%)

The patient was discharged after the 5th post-operative day. Thirty four months post hepatectomy, the patient presented with symptomatic lung metastasis treated by sorafenib. She died 50 months after liver resection and 16 months after the diagnosis of lung metastasis.

DISCUSSION

The most common location of the ACC remains the salivary glands. They represent 10% of salivary carcinomas [1, 6]. Isolated hepatic metastases have been rarely reported; they often present with multiple visceral metastases where only a medical treatment is possible [7]. For the best of our knowledge, we report the 9th liver resection for ACC metastasis published in English literature.

The evolution of these tumors is difficult to predict. Poor prognosis factors are the tumor size, the site of the primitive and the existence of metastases [8]. The index of tumor proliferation Ki- 67 could also have a prognosis value [9].

Given the rarity of these tumors, the studies reporting series of non-colorectal non-endocrine liver metastases didn't identify any case of hepatic metastasis of ACC [10]. In the multicenter series of

Adam et al [11] which include 1452 patients, only 15 patients had liver metastases of a primitive head and neck tumor. The main histological type in this patients' sub-group was the epidermoid carcinoma and the 05 years survival rate was less than 15%. In our patient, the diagnosis of an ACC was done at the age of 63 and the hepatic metastasis appeared 46 months after the resection of the primitive tumor.

The therapeutic strategy is not clearly defined [6]. In the literature, few cases of resection of liver metastasis of ACC have been reported. Zeidan et al [5], Sali et al [12] and Park et al. [13] reported a case of hepatic resection for ACC metastasis; the primitive was respectively a lacrimal ACC, parotid or sub-maxillary ACC and Tracheal ACC [5, 12, 13]. All published cases in the English literature are summarized in Table I. Authors were contacted by e-mail to update the patients' observations.

Table I: Summary of published cases with resection of hepatic metastasis of ACC

Authors	Age	Sex	Primary Tm	Metastasis	Delay tm- Mt	Surgery	Medical treatment	Survival
Zeidan 2006 [5]	51	M	Lacrymal gland	Right liver (20 cm)	20 y	Right hemi hepatectomy	None	Lost to follow-up
Park 2009 [13]	52	F	Trachea	Segments VIII, VII	36 m	Right hemi hepatectomy	None	
Balducci 2011 [7]	55	M	Sub- maxillary	Segments VIII, V, VI	18 m	Right extended hepatectomy	Cisplatin Epirubicin	Died after 24 m -Hepatic recurrence Sgt VI after 21m: Metastasectomy.
Scuderi 2011 [7]	30	F	Parotid gland	Segment III	11 y	Left lobectomy	None	-After 10m, bilateral lung mt and brain damage -Died 03 m later
Coupland 2014 [6]	52	F	Sub- maxillary	Segment V	38 y	sub segmentectomy V	None	02 y alive
Spolverato 2014 [1]	59	F	Sub- maxillary	IV	24 m	Left hemi hepatectomy	None	05 m alive
Sali 2016 [12]	42	F	Lacrymal gland	Segments IVb & VII Lung Mt	60 m	-Right hemi hepatectomy extended to segment IV b -Lung metastasis resection	None	18 m alive after lung surgery
Zemni I [14]	29	F	Parotid gland	Left liver	60 m	Wedge resection	None	Alive
Present Case	63	F	Lacrymal gland	Left liver	46 m	Left hemi hepatectomy	Sorafenib after lung metastasis	recurrence at 26 m (lung metastasis); Patient died after 50 m of follow-up

M: Male; F: Female; Mt: metastasis; Tm: Tumor; m: months; y: years.

Considering the slow evolution of these tumors, many questions raised the benefit of the surgery and/or chemotherapy in this context especially that the metastases are often asymptomatic [14-16]. For the isolated liver metastasis, especially with free gap between the primitive and the metastasis, the surgery of resection was performed. Interesting survivals were reported in the published case reports which suggest the benefit of a surgical resection. Scuderi et al [8] reported re- hepatectomy after a unique metastatic recurrence. laparoscopic approach could be considered.

More free the gap between the primitive and the metastasis is important, better the prognosis is and the resection of the hepatic metastasis is justified. In the reported cases, the free gap ranged from 18 months to 38 years [1, 5-8, 13]. For our patient, the free gap between the primitive and the hepatic metastasis was 46 months. The definitive diagnosis lays on the histological study of the operative specimen using immunohistochemistry. Spolverato et al [1] reported a positivity in the cytokeratin 7, c-Kit, and EMA and a negativity for the PAX8, TTF-1, CDX2, CK20, chromogranin,

synaptophysin, CD5, Hepar 1, the ER, and the p 63.

A neo adjuvant chemotherapy with an association of pirubicin and cisplatin was suggested by Balducci and al [7]. The response to this protocol was poor and a hepatic resection was carried out afterwards. An adjuvant chemotherapy was also proposed by some authors but the benefit was not clearly established [5, 6]. Intra Arterial Cytoreductive Chemotherapy is now being considered as an adjuvant therapy of ACC to improve disease control after a primary surgical resection [12].

The prognosis after resection remains favorable. However, the fact that publications of cases of resection of hepatic ACC metastases are rare makes it that we cannot estimate the actual prognosis of this presentation.

CONCLUSION

Hepatic metastases of the ACC are rare. Surgery is proposed when the metastasis is isolated, a fortiori when there is a free gap between the primary tumor and the metastasis.

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CONFLICTS OF INTEREST:

The authors declare no conflict of interest

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