



Case report

A rare case of hepatic metastasis 20 years after surgical resection of a thymoma: A case report

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ABSTRACT

Introduction: Thymoma is the most common solid, primary mediastinal tumour, accounting for 20% of mediastinal neoplasms. Malignant thymus tumours (thymomas and thymic carcinomas) metastasize primarily locally. Distant metastases, especially to liver are very rare.

Presentation of case: We review the case of a 59 year-old female patient, who underwent resection of the thymus with a diagnosed type B2 thymoma 20 years ago. The patient was referred to our hospital with a newly discovered space-occupying lesion in the liver, which had been detected in a routine follow-up magnetic resonance imaging scan. Since a malignant tumour of the liver could not be excluded, a conventional left hemihepatectomy was performed. Histological examination revealed a liver metastasis of the type B2 thymoma, which had been removed 20 years ago.

Clinical discussion: The case was discussed in the interdisciplinary tumour board. Based on the very long history of the primary removal of the thymoma as well as the R0 resection of the liver metastasis, a follow-up regimen with CT scans on a regular basis was recommended.

Conclusions: Newly discovered lesions of the liver in patients even with a long history of a thymoma should raise the suspicion of a liver metastasis that should be surgically resected as the therapy of choice. Further, this case indicates the importance for long-term radiographic follow-up.

1. Introduction

Thymoma is a tumour that originates from thymic epithelium or cells with thymic epithelial differentiation [1]. The incidence of thymoma in Europe constitutes about 1.7 million cases per year [2] which indicates that thymomas are rare mediastinal tumours. However, of all space-occupying lesions in the anterior mediastinum, thymomas are responsible for about 50% of all cases [4]. Approximately 10–15% of thymomas are associated with myasthenia gravis [17]. Most thymus tumours grow slowly and predominantly invade surrounding structures, whereas distant metastasis in the liver, bones, lymph nodes or lungs are described to be rare [5,6,7].

Thymomas are classified according to the World Health Organisation (WHO) into Type A, AB, B1, B2, B3 and thymic carcinoma (former called type C) [8]. About 75% of thymomas are classified as benign and 25% evolve into malignant, so-called thymic carcinoma [3]. Metastasis occurs primarily to the pleural cavity, lungs or bones [9]. Extrathoracic

metastases of thymus tumours are extremely rare and only described in a few cases in the literature. In 1983, Ichino et al. were the first to describe and document 83 cases of extrathoracic metastases, noting that a histological origin has not been validated [18]. Follow-up studies by this group summarized liver, bone and lymph nodes as common sites [9]. In particular, liver metastases often remain asymptomatic and present as space-occupying lesions of the liver, usually diagnosed incidentally during routine or follow-up examinations or due to pain in the right upper quadrant of the abdomen. Very rarely liver metastases display as haemoperitoneum or liver rupture [10,11]. The therapeutic approach of liver metastasis depends on the type of the thymic tumour and whether metastatic status is considered metachronous or synchronous. Synchronous liver metastases are treated using adjuvant chemotherapy and surgical resection of both thymic tumour and liver metastasis. Metachronous liver metastases require surgical resection and chemotherapy as an option in selected cases. The use of platinum-based chemotherapy in the treatment of liver metastasis of thymic tumours has been shown to

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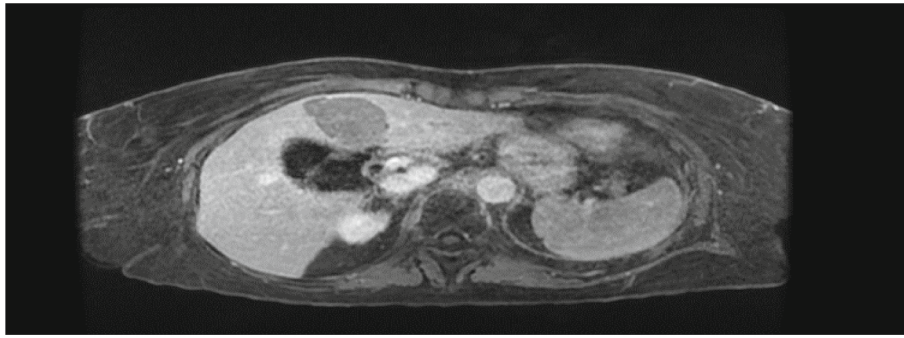
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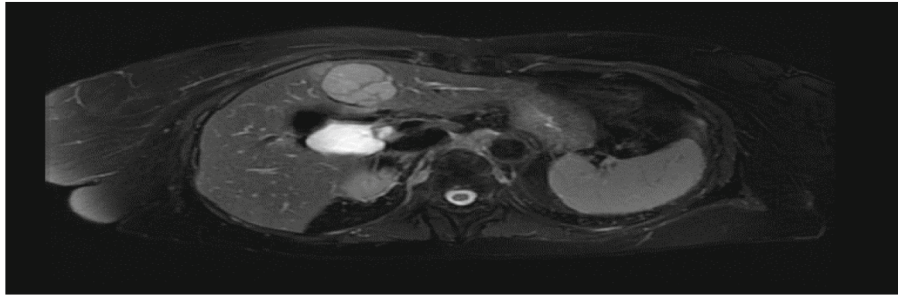
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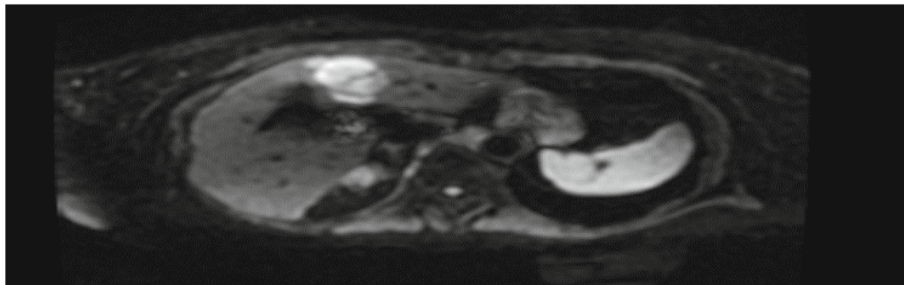
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Picture 1



Picture 2



Picture 3

Fig. 1. Pictures 1,2,3 MRI of the abdomen showing a subcapsular space occupying lesion in segment IVb of the liver.

have a beneficial effect on survival rate of patients [12,13]. Only few studies to-date have addressed treatment regimens such as resection or radiotherapy of the metastasis in patients with distant metastasis [14,15].

The presented patient was treated in the surgical department in Siloah Hospital in Hannover, Germany. The work of this case report has been reported in line with the SCARE criteria [19]. A written informed consent was obtained from the patient for publication of this case report. The patient received detailed information about the therapeutic strategy and operative procedure.

2. Case presentation

A 59 year old female patient was presented with a newly discovered space-occupying lesion of the liver in May 2020. The lesion was detected incidentally during a routine follow-up CT and MRI scan 20 years after resection of a primary type B 2 thymoma and about 18 years after resection of a recurrent tumour in the pleural cavity. Both radiological examinations (CT and MRI) showed a lesion of 4–5 cm in diameter located primarily in liver segment IVb (Fig. 1). The appearance of the lesion was suspicious for either an adenoma or focal nodular hyperplasia (FNH) of the liver. Other differential diagnoses such as echinococcal disease (hydatid disease) were ruled out as the patient underwent an operation for this condition in 2008.

Since the morphology in particular of this newly discovered space-occupying lesion of the liver suggested a malignant origin, a liver operation was indicated. Intraoperatively we found a 5 cm large tumour mainly in segment IVb that reached into segment IVa and segment III of the liver. After complete mobilisation of the liver, an intraoperative ultrasound examination was performed that revealed no further lesions in the liver. Because of the tumour size and the intrahepatic localisation, we performed a conventional left-sided hemihepatectomy, resecting segments II-IV, respectively. First the left hepatic artery and the left portal vein were dissected followed by the dissection of the left hepatic vein. The dissection of the liver parenchyma was carried out using the Cusa Device combined with a Pringle manoeuvre for about 15 min. There was no relevant blood loss (less than 200 ml) and thus there was no need for blood transfusion intraoperatively. The Procedure was performed by a surgeon with more than 20 years of experience in hepatobiliary surgery. The postoperative course was without any complications. The peak levels of liver enzymes (AST/ALT) were reached on the 5th postoperative day. There was no need for a postoperative substitution of clotting factors. The patient was discharged home on the 11th postoperative day in good health. Unexpectedly, subsequent pathological examination revealed type B2 thymoma cells, leading to the conclusion that the resected tumour was a liver metastasis originating from a type B thymoma, which had been resected 20 years ago (Fig. 2).

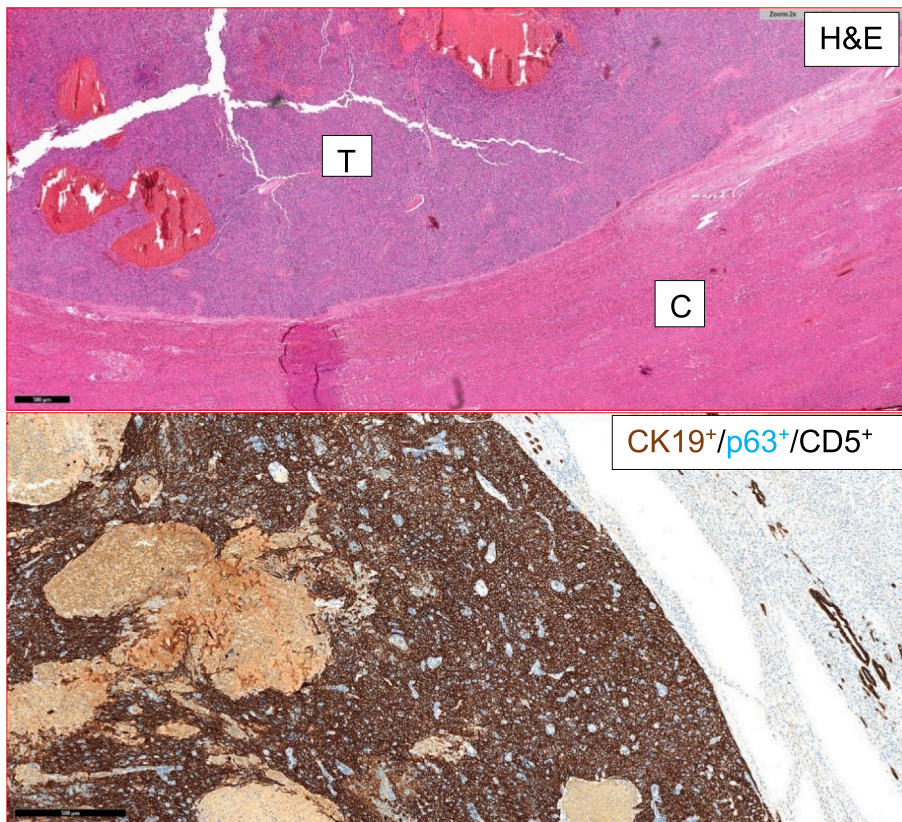


Fig. 2. The upper picture displays immunohistological staining of the liver showing the malignant lesion (T) surrounded by a capsule of connective tissue (C). (The capsule contains cells with eosinophilic cytoplasm). The lower picture shows anti-CK19 (brown), anti-p63 and anti-CD5 staining of tumour cells. These cells show a negative staining-reaction against antigen Pax8 und CD20, which is a characteristic constellation of findings for thymoma type B2. (For interpretation of the references to colour in this figure legend, the reader is referred to the web version of this article.)

3. Discussion

Thymoma is the most common solid, primary mediastinal tumour, accounting for 20% of mediastinal neoplasms. 90% of thymoma occurs in the anterior superior mediastinum, and a smaller percentage occurs in the neck and posterior mediastinum or other localisations [16]. Most thymus tumours grow slowly and predominantly invade surrounding structures, whereas distant metastasis in the liver, bone, lymph nodes or lungs are described to be rare [5,6,7].

There are very few published studies demonstrating liver metastases of a thymoma. Usually, liver metastasis occurs within the first 5 years after the primary diagnosis. Here, we report a patient who presented with liver metastasis 20 years after primary resection of a mediastinal type B2 thymoma. Only two other documented cases exist in the literature which report liver metastases, 13 years and 22 years after primary resection, respectively. The treatment of liver metastasis consists of surgical resection, potentially followed by chemotherapy with optional radiation therapy.

Our presented case was discussed in the multidisciplinary tumour board. Considering the long history of 20 years after primary resection of the thymoma and zero residual tumour after liver metastasis resection, no chemotherapy was recommended. However, regular follow-up CT scans every 6 months should be performed.

For patients with liver metastases long after resection of the primary tumour, curative liver operation should be performed whenever technically feasible as the treatment of choice; other modalities are less favorable. There are few data on long term survival after resection of liver metastases from thymoma primaries. An important implication from this case is that long term monitoring, even more than 20 years, should be implemented on a regular basis.

4. Conclusion

Thymomas are rare tumours of the anterior mediastinum, which

rarely metastasize to the liver. This case describes an atypical late metastasis of a thymoma in the liver 20 years after the primary removal of the thymoma type B 2 in the mediastinum. It underlines the importance that all newly discovered lesions of the liver in patients with a history of a thymoma should raise the suspicion of a liver metastasis independently of the first surgical resection of the primary tumour.

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Ethical approval

Not required.

Consent

A written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Guarantor

Fetyan Dhahir Ali

CRedit authorship contribution statement

Fetyan Dhahir Ali: Main author, data collection, data analysis, writing the paper,

Stefan Kuebler: data collection.

Laura Hermann: Patient care and data analysis.

Nora Lakenberg: data review und grammar check.

Julian Mall: data analysis and interpretation.

Josef Fangmann: study concept and design, supervising the whole process of writing the case report, performed the operation.

Declaration of competing interest

None.

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