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RARE CASE OF UNANTICIPATED HEPATIC TISSUE IN THE THORAX MIMICKING A PULMONARY MASS IN A 49-YEAR OLD FEMALE

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Abstract
Accessory liver tissue (ALT) is a rare congenital anomaly, usually inherited in an autosomal recessive pattern, though rarely reported after diaphragmatic trauma or surgery. Approximately 23 cases of ectopic liver tissue have been described. The gallbladder is the most common site; intrathoracic accessory liver lobe is an extremely rare variant, characterized by autonomous hepatic tissue within the thoracic cavity. Its clinical significance remains unclear, but malignant transformation has been suggested, possibly due to chronic inflammation from bile and venous stasis. No specific diagnostic or management guidelines exist, and most cases are diagnosed retrospectively following surgery. A 49-year-old woman with a 24-year history of tobacco chewing and family history of malignancy presented to a tobacco cessation clinic. Initially asymptomatic, she later reported left cheek pain radiating to the ear and neck. Clinical examination was unremarkable, with no intraoral lesions. Imaging was performed to exclude malignancy. Contrast-enhanced CT thorax incidentally revealed a well-defined, heterogeneously enhancing pleural-based lesion (53.5 × 52.6 mm) in the right lower lobe, without significant lymphadenopathy. Given her risk factors, CT-guided biopsy was undertaken. Histopathology showed benign hepatocytes arranged in cords and plates with intervening sinusoids, mild macrovesicular steatosis, and periportal lymphocytic infiltrates. No malignancy or lung parenchyma was identified. Findings were confirmed as benign hepatic tissue, establishing the diagnosis of intrathoracic accessory liver lobe. Serum alpha-fetoprotein was normal. As the patient remains asymptomatic, six-monthly follow-up has been planned. This rare case underscores the importance of considering anatomical aberrations in evaluating suspicious thoracic masses, guiding appropriate management and surveillance.

Keywords: *hepatocellular carcinoma, incidental radiological finding, malignant transformation, serum alpha-feto protein, accessory lobe of liver, ectopic liver tissue, intrathoracic accessory lobe of liver.*

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INTRODUCTION

Accessory Lobe of the Liver (ALL) and heterotopic liver tissue are rare entities, with reported incidences of 0.5% and 0.1%, respectively [1]. Intrathoracic accessory liver lobe is an uncommon variant in which ectopic hepatic parenchyma is located within the thoracic cavity. It may be congenital or acquired following thoracic trauma or surgery [2]. Although reported from the neonatal period to 75 years of age and more frequently in females, its exact etiology remains unclear. Owing to its rarity, only limited cases are documented in the

literature. Clinically, intrathoracic ALL is usually asymptomatic and detected incidentally on imaging or during thoracic surgery. Symptomatic cases may result from compression of adjacent structures, presenting with cough, dyspnea, chest or back pain, and rarely dysphagia or indigestion. Management is guided by symptoms. Asymptomatic patients without complications may be managed conservatively with periodic follow-up. However, many cases are misdiagnosed as pulmonary tumors, sequestration, or hydatid cysts, leading to unnecessary thoracotomy. In contrast, pedunculated abdominal ALL carries a risk of torsion, infarction, or rupture, often requiring emergency surgery.

Ectopic liver tissue is thought to have increased malignant potential due to compromised vascular and biliary drainage. Morita et al. [3] reported 39 cases of hepatocellular carcinoma arising in heterotopic liver tissue, predominantly in the abdomen, with only one intrathoracic case. Multidisciplinary evaluation is essential for accurate diagnosis and management.

CASE PRESENTATION

A 49-year-old woman with diabetes, hypothyroidism, prior acute kidney injury, a 24-year history of tobacco chewing, and family history of malignancy presented to our tobacco cessation clinic. Initially asymptomatic, she later developed persistent left cheek pain radiating to the ear and neck. Examination revealed a 1×1 cm left upper deep cervical lymph node and poor oral hygiene without intraoral lesions. CT thorax revealed a well-defined, heterogeneously enhancing pleural-based mass (53.5 × 52.6 mm) in the posterior and medial basal segments of the right lower lobe, displacing pulmonary vessels, without significant lymphadenopathy.

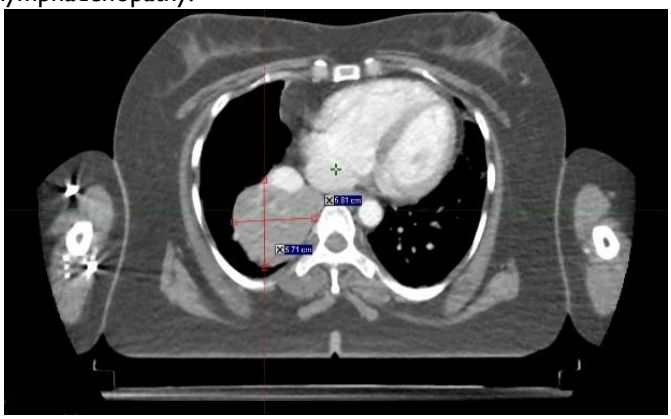

Figure 01: Contrast enhanced CT Thorax (Axial view) - Evidence of a well circumscribed lesion measuring 5.7x5.8cm in right hemi-thorax.

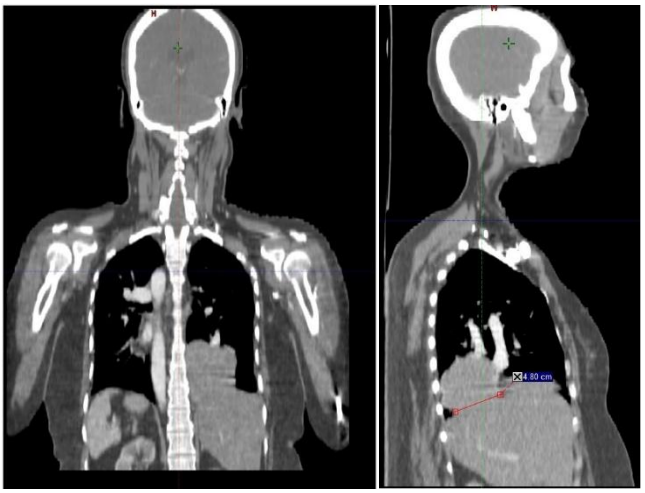

Figure 02: Contrast enhanced CT Thorax (Coronal and Sagittal view) demonstrating lesion in right hemi-thorax extending via the defect in right diaphragm measuring 4.7cm. Lesion arises from liver and is supplied by branch of left hepatic artery, as well as drained by left hepatic vein

This pleural based lesion was an incidental finding. In view if her tobacco use, family history and other risk factors, we planned a CT-guided biopsy of the lung mass to rule out malignancy. Histopathological examination of the biopsy revealed hepatic parenchyma composed of benign hepatocytes arranged in cords and plates with intervening sinusoids. The hepatocytes exhibited mild macro vesicular steatosis and mild peri-portal lymphocytic infiltrates. There has been no evidence of malignancy and no lung parenchyma in specimen, and deeper sections confirmed these findings. The impression was given as benign hepatic parenchyma with steatosis. The findings were further discussed with the pathologist, who confirmed that the biopsy, taken from the lung mass, consisted solely of liver tissue.

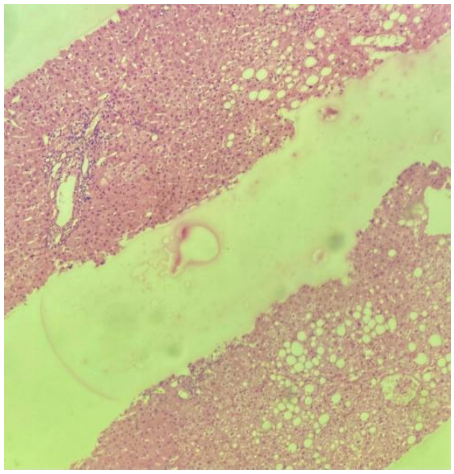

Figure 03: Core biopsy of liver showing preserved architecture and steatotic areas (H&E, x100)

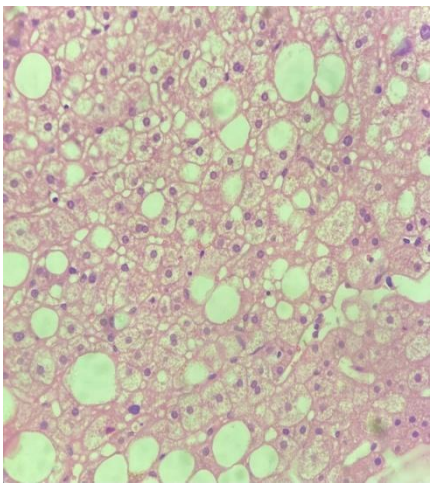

Figure 04: Liver biopsy showing macrovesicular steatosis. The hepatocyte cytoplasm is distended by lipid vacuoles displacing the nucleus peripherally (H&E, x400)

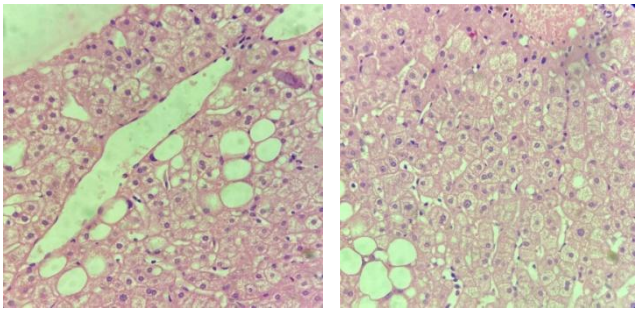

Figure 05A and 05B: Hepatocytes exhibiting both microvesicular and macrovesicular steatosis (H&E, x400)

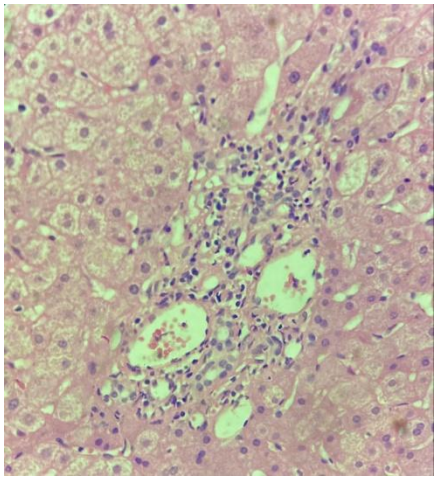

Figure 06: Portal tract showing mild chronic inflammatory infiltrate composed predominantly of lymphocytes and few plasma cells. Bile pigment is seen within few hepatocytes (H&E, x400)

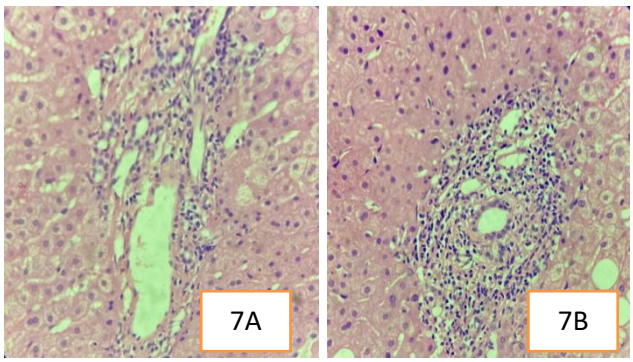

Figure 07A and 07B: Portal- based inflammation (H&E, x400)

At present, the patient remains stable and asymptomatic with respect to the accessory liver lobe in the lung. Tumour markers like serum Alpha Fetoprotein and Serum Carcino Embryonic Antigen done at 3 months were within normal limits. She is planned to be kept under follow-up, with imaging after three months to look for any suspicious changes. As our patient was asymptomatic and there was no suspicion of underlying malignancy in the ALL, she was kept on follow-up with imaging and serum alpha fetoprotein.

DISCUSSION

Historically, accessory lobe of liver (ALL) has been reported as incidental finding during surgery or at autopsy. The first case of accessory liver tissue was reported in 1957 by Hansbrough and Lipin. Thereafter, only three cases of ALL were reported every decade from 1925 to 2006. Till date, only around 23 cases of ectopic liver tissue have been reported, most are isolated case reports or series. This results in serious dilemmas in terms of management, with no specific guidelines available. Most cases of intrathoracic ALL end up in surgery and are diagnosed retrospectively. Hence, it is very important to consider intrathoracic ALL as a possible diagnosis in any patient presenting with lung mass to avoid unnecessary surgery.

Accessory liver tissue is a very rare anatomical abnormality and mostly results from embryonic heteroplasia, though in rare instances it has been reported after diaphragmatic trauma or surgery. The rarity of this condition is attributed to its autosomal recessive mode of inheritance. During the fourth week of development, the hepatobiliary system develops from the hepatic diverticulum as a ventral outgrowth of foregut. There are two hypotheses regarding ALL, one claims that the embryonic liver curls outwards resulting in ectopic liver tissue, while other states it is a result of increased intra-abdominal pressure due to the rapidly developing liver [4]. Accessory liver tissue may prevent closing of the umbilical ring, which occurs in 7th week of gestation, and hence is commonly associated with acromphalus.

The most common site of occurrence is in the gallbladder. Akura et al reported 76 cases pf ALL, all occurring in the abdominal cavity. The most prevalent site (65 percent cases)

was in and around the gallbladder [5,6]. Other reported sites were in the pancreas, spleen, adrenal glands, retroperitoneum, or omentum or pelvis. Stattaus et al classified accessory liver tissue in 2 ways: accessory lobe with connection to liver (pedunculated ALL or sessile ALL) or completely separate accessory lobe (ectopic ALL). Further, there are several types based on the volume and weight of Accessory liver tissue. There is no need for surgery in patients with sessile ALL or ectopic ALL in the absence of symptoms. However, for pedunculated ALL, surgery should be performed to avoid complications like torsion.

Abdominal ALL are commonly misdiagnosed as enlarged periportal lymph nodes or intraperitoneal tumour. If present in the thorax, they are mistaken for pulmonary or diaphragm tumours. Advances in imaging, such as endoscopic ultrasound, CT, MRI (Magnetic Resonance Imaging), PET (Positron Emission Tomography), and multislice spiral CT (MSCT) and multiplane MRI imaging, are of immense help in providing more accurate information for diagnosis [7].

Wang et al [8] (2010) reported a 39-year-old man who underwent thoracotomy for a suspected lung tumour. However, intraoperatively, there was a well circumscribed mass connected to liver by small pedicle passing via the diaphragm. In retrospect, the diagnosis of ALL could have been diagnosed if only appropriate radiologic investigations had been performed. A complete evaluation using multiple imaging techniques including radiographs, CT, MRI, radioactive imaging, or type-B ultrasound, is necessary.

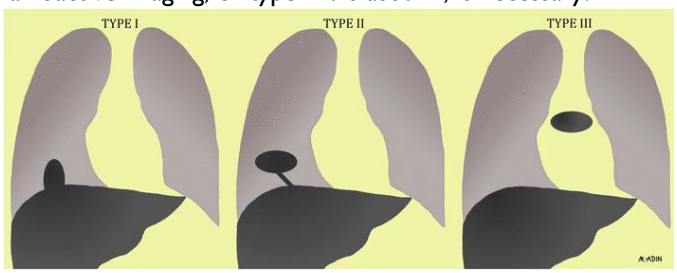

Figure 08: Classification of intra-thoracic accessory liver tissue: Type 1: Direct extension of liver tissue into the thoracic cavity through a diaphragmatic defect. Type 2: An accessory liver within the thoracic cavity. Type 3: Liver tissue located in thoracic cavity, entirely independent of the intra-abdominal liver (ectopic liver) [9].

Some radiological features helpful to differentiate Accessory Liver tissue (ALL) from tumours are, the density of the mass may sometimes differ from normal hepatic parenchyma, or presence of a connecting pedicle. In addition, multidetector spiral CT scan, along with multi-planar reconstruction and MRI, can reveal the exact location of mass in relation to normal liver parenchyma. Further, in hepatic angiography if there are vessels extending to the mass, it is suggestive of ALL. Endoscopic ultrasound and thoracoscopy also aids in diagnosis of intrathoracic ALL. Many patients have no clinical signs or symptoms in spite of a large mass. On imaging, the mass pushed the surrounding organs without frank invasion. This is different from an invasive malignant tumour that invades surrounding tissues and metastasizes to distant organs. Differentiating a completely separate intrathoracic ALL from benign tumour is quite challenging without pathological confirmation.

Ball et al [7] reported 28-year-old woman presenting with sudden-onset stabbing chest pain as well as dyspnea. CT angiogram revealed a 2.2 x 2 cm mass, adjacent to and possibly arising from left anterolateral wall of oesophagus. Differential diagnoses considered for a posterior mediastinal mass were neurogenic tumours, lymphoma, and, less commonly, oesophageal cancer. However, endoscopic ultrasound revealed a hypoechoic mass with connection to liver, raising the possibility of intrathoracic ALL. Single Photon Emission Computed Tomography - Computed Tomography (SPECT-CT) demonstrated a transdiaphragmatic stalk confirming diagnosis of intrathoracic ALL.

Wang et al [8] reported three cases of accessory lobes of liver. The first case was a pedunculated ALL, the second case was true ectopic liver, as well as third case a sessile ALL. All three cases have been diagnosed by CT or MRI findings. CT and MRI findings from three cases were: (i) a substantial part of accessory lobe had same signal density as that of normal liver tissue; (ii) ALL was distinct with smooth margins and was well demarcation; (iii) presence of stalk connecting to the liver (iv) flow-void vascular imaging apparent in MRI. Diagnosing a intrathoracic ALL is difficult since it is completely separate and cannot be readily differentiated from a mediastinal mass but appropriate use of imaging modalities can aid in accurate diagnosis. Diagnosis of ALL is essential to avoid unnecessary surgery, and as the ectopic tissue is at a higher risk of malignant transformation, and to be kept under follow up.

Until 1985, only seventy cases of ectopic liver have been reported in literature, according to Yamashita et al [9]. Of these 70 patients, nine patients eventually developed hepatocellular carcinoma in the ectopic liver. Additionally, one patient developed hepatocellular carcinoma in mother's liver but not in accessory liver lobe. Arakawa et al [10] reviewed 21 patients with hepatocellular carcinoma (HCC) that developed in ectopic liver tissue with mother liver being tumour-free. Though there have been a few occurrences in Caucasians, most cases of hepatocellular carcinoma occurring in ectopic or auxiliary liver are seen in South-East Asians [10]. The reason for malignant transformation in ALL may be the altered vascular outflow or bile stasis due to ineffective bile reabsorption. There aren't many reports of ALL being linked to benign conditions such as focal nodular hyperplasia. Arakawa et al [10] reported that in 12 out of 21 instances (around 63%) of HCC developing in ectopic liver tissue, serum alpha-fetoprotein level was greater than 1000 ng/mL. Hence, it is necessary to consider possibility of HCC in an accessory liver tissue, when there is serial increase in serum AFP without any discernible pathology in the native liver. To our knowledge, there is only one patient treated by laparoscopic surgery and embolization of the feeding artery for a HCC developing in ALL.

Polikarpova et al [11] reported a 72-year-old man who had been admitted under orthopaedics with left ankle injury

following a fall. Routine chest X-ray incidentally showed a right lower lobe chest mass. CT chest scan showed a well-defined lesion seemingly originating from the liver via a defect in the diaphragm. As serum alpha-fetoprotein level was only 1 kIU/L and the imaging did not point to any underlying malignancy, a conservative approach was adopted, and he was kept on follow-up every six months. The patient did not have any further issues and remained in good health. There is no evidence to propose that six monthly yearly follow-up is superior to annual surveillance [11]. Therefore, the decision is based on clinical judgment in case-to-case basis.

CONCLUSION

In conclusion, intrathoracic accessory liver tissue is usually incidentally detected while evaluating for unrelated medical conditions. This highlights the importance of comprehensive diagnostic tests that can differentiate between anatomical aberrations and pathologies. Malignant transformation in accessory liver tissue is the main cause for concern. Annual surveillance with liver function tests, chest and abdominal CT imaging, and specialist follow-up is advocated. Study of intrathoracic accessory liver tissue highlights the remarkable complexity and diversity of the human body. Despite being an extremely rare variant, knowledge of this trait and its clinical implications will help in better diagnosis and treatment for those affected. Basic research into the underlying mechanisms of embryogenesis and the origin of ALL, as well as genetic predispositions and long-term consequences of ALL if needed, to bridge the void in our current knowledge about this condition. Our case report is a rare diagnosis of incidentally detected intrathoracic accessory liver tissue, that is most likely result of congenital malformation. Management depends on symptoms, anatomy of accessory lobe, and clinical and biochemical suspicion of underlying malignancy. Understanding of this rare condition is pivotal for prompt diagnosis of this infrequent condition and appropriate management and surveillance.

DISCLOSURES

Informed consent for treatment and open access publication was obtained or waived by all participants in this study. Conflicts of interest: In compliance with the ICMJE uniform disclosure form, all authors declare the following: All authors have declared that no financial support was received from any organization for the submitted work. All authors have declared that they have no financial relationships at present or within the previous three years with any organizations that might have an interest in the submitted work.

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CONFLICT OF INTEREST

Authors are declared that no conflict of interest

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INFORM CONSENT AND ETHICAL CONSIDERATIONS

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