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INFLAMMATORY FIBROBLAST TUMOR AND LIVER ABSCESSES IN THE YOUNG PATIENT

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ABSTRACT

Introduction: Inflammatory fibroblast tumor is rare tumor that most often occurs in younger people, usually 30 years old or younger, but most commonly in children age 6-10 years. It usually affects gastrointestinal tract and the lungs but it can also occur in several places at the same time. Clinical manifestations vary depending of the affected system of the body so it is very difficult to determine diagnosis without surgical extirpation and patohistological analysis. Complete surgical resection is curative in most patients and recidivism is rare. Liver abscesses more common occur in females with risk factors and medical history of diabetes, previous liver disease and less likely in patients with granulomatous diseases. Liver abscesses mortality in developing countries is 2-12%, increasing due to open surgical drainage.

Case report: We present a 35 years-old patient who was treated at the Clinic for Infectious Diseases University Hospital Mostar and University Hospital Sarajevo, Clinic for Infectious Diseases in August and September 2018 and Clinical Hospital Merkur, Zagreb Surgery Clinic in December 2018. Data was used from medical documentation. Young, immunocompetent patient who was addmited to a hospital following high fever, chills and poor general condition was diagnosed with multiple focal necrotic lesions, differential-diagnostically most likely liver and spleen abscesses with high suspicion of liver malignancy. Liver biopsy was performed and patohistological analysis confirmed the diagnosis of multiple liver abscesses in the IV and VI liver segment, and inflamatory fibroblast tumor in the IV liver segment. Eight weeks of conservative treatment resulted in a complete regression of liver abscesses and inflamatory fibroblast tumor was surgically extirpated at the Clinic Hospital Merkur, Surgery Clinic in the Zagreb in December 2018.

Conclusion: An approach to a patient with a multiple liver abscesses and liver tumor requires sub-specialists experience and urgent multidisciplinary diagnostic and treatment approach to prevent further complications and deadly outcome.

Key words: inflammatory fibroblast tumor, liver abscess, antibiotics

INTRODUCTION

Inflammatory Myofibroblastine Tumor (IMT) is a rare tumor in younger people, up to 30 years of age, but more often in children and most commonly affects the gastrointestinal tract and lungs. Clinical manifestations depends on the affected system but in most cases it is asymptomatic. It is very difficult to set the diagnosis without surgical extirpation and patohistological analysis. Complete surgical resection means cure for most patients. IMT is primary benign liver tumor and in literature is described under different names as a plasma cells granuloma or inflammatory pseudotumor [1]. In most cases it is found randomly as a result of wide use of imaging but the etiology and the nature of this condition remains unknown, and the therapeutical approach is still unclear and controversial [2-4].

Gastroenerologist and hepatologist consultation is often needed [3]. European reports confirm the disease more often in younger women, and the first major study of this entity in Japan shows higher prevalence in men any age and in both sexes but with an average lesion size less than in previous European reports [5]. Piogenic liver abscesses are relatively rare disease [6]. According to etiology, liver abscesses can be bacterial (pyogenic) and parasitic (ex. *Echinococcus granulosus*

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liver abscess). Main clinical manifestation is fever of unknown origin [7]. Liver abscesses more common occur in females with risk factors and medical history of diabetes, previous liver disease and less likely in patients with granulomatous diseases. Liver abscesses mortality in developing countries is 2-12%, increasing due to open surgical drainage [8-11].

Conservative treatment recommendation is a combined antimicrobial therapy with meropenem, vancomycin, metronidazole (during 4 weeks) and fluconazole (during 20 days). Active treatment is usually classical surgical approach or laparoscopy, in a specific cases percutaneus approach can be performed [7]. Pyogenic liver abscess is a challenging disease with a high rate of postoperative morbidity. Most abscesses are located in the right liver lobe. Imaging techniques especially abdominal ultrasound and MSCT are essential for the diagnosis and treatment of the liver abscesses [7].

CASE REPORT

Previously healthy immunocompetent patient age 35, engineer and the father of two children, was addmited to our hospital following high fever up to 39.4C with chills, strong headache, nausea, vomiting and poor general condition. His symptoms were in progression two days before hospitalisation. Two months earlier he noticed shortness of breath even after simple every day activities. In the past he had a history of hem

Family history revealed that his father was treated in the last two months because of the gastric cancer. Epidemiology anamnesis showed that he had contacts with rabbits and hens, and that he was previously on a vacation in the Adriatic coast with his family.

During the examination he was febrile (38.5C), his blood pressure was 110/80mmHg, RF 19/min, SO_2 95% and c/p 115/min, GCS 15/15, BW 115kg, his skin was pale and the rest of the physical status including heart, lungs and abdomen was normal.

Laboratory tests inicially revealed elevated inflammatory parameters which were in regression during treatment (SE 84-93 mm / 3.6 h, CRP 307-150 mg / L, L 21,6-10,2, DBC neutrophils 86% ly 7-8% eo 2%, T 355-801, AST 61-249 U/L, ALT 83-262 U/L, GGT 233-225U / L, LDH 184-277 U / L, and other findings including red blood count, kidney parameters, glucose, electrolytes, coagulogram, acid status and urine were in the referent value.

The results of the main radiological and microbiological at the addmision at the Clinic for Infectious Diseases Clinical Hospital Mostar are showed in the Table 1.

Table 1. Results of microbiological and radiological exminations at the Clinic for Infectious Diseases University Hospital Mostar

Blood culture	negative
Urin culture	sterile
Rose Bengal test	negative
Ophtalmological examination (FUO)	PNO with clear boundaries, in the level of the surrounding retina. Vascular and maculas are neat.
CSF	Normal result
Chest radiography	Normal result
Ultrasound of the abdomen	Liver lesion along fissura lig. differential diagnostically is FNH or neoplasm. Spleen lesion can be also abscess but it can also be other etiology.
MSCT abdomen	Necrotizing lesion is visible in S8 about 7,2x6,7 cm in the longest diameter, ventral in S4 about 5,5x4,8 cm, low in S6 about 5 cm in diameter with a central necrotic part about 2,8 cm. Lesions do not contain air. Hypervascular lesion in S4 is about 6x6.3 cm described as a possible adenoma, FNH or HCC. Necrotizing spleen lesions are demarcated and 3.2 cm. Picture 1 and 2MSCT abdominal liver
Colonoscopy	Mucouse membrane is intact, ampoules recti vein splet gr. II.
Control MSCT of the liver two months later	Previously described tm formation in S4 is 6 cm is most likely hemangioma or FNH. Described abscesses in the liver are now smaller in size without visible capsule, size 4 cm in S6 and 3cm in S5.

Considering that the diagnostic tests revealed multiple focal necrotic lesions of the liver and spleen with a high suspicion of abscess or even tumor, multidisciplinary approach was required. A gastroenterologist, surgeon and a radiologist were involved during the whole procedure of treatment. Gastroenterologists consilium found that multiple abscesses required urgent surgical treatment according to gastroenterological algorhitms. Surgical consilium found that surgical treatment wasnt an option in that moment because of the unclear etiology of the liver lesions and an acute infection. They recommended a needle biopsy of the lesions.

Since the requested radiological procedures (MR of the liver) and the needle liver biopsy couldnt be performed in our hospital at the time the patient was sent to a Clinic for infectious diseases in Sarajevo for further evaluation. After 13 days of conservative treatment his inflammatory results were in regression but he was still febrile with the elevation of the liver transaminases.

Antimicrobial empiric parenteral therapy with ceftriaxone was administered initially, but after the radiological abdominal evaluation was replaced with meropenem, clindamycin and metronidazole p.e. (11 days). In Sarajevo hospital, general microbiological and diagnostic procedure was performed and is presented in Table 2.

Table 2. Results of microbiological and radiological exminations at the Clinic for Infectious Diseases University Clinical Center Sarajevo

Blood culture	Negative
Urin culture	Sterile
Cardiologist opinion and heart ultrasound	Normal result
Ultrasound of the abdomen	Described liver changes may be responsive to an organized CT verified abscess. A change in the liver along the ligamentary fission area is like a etiologic high nodular hyperplasia, neo process. Promise in the spleen corresponds to abscess, but no other etiology is excluded
MSCT abdomen	Multiple liver leasions are most likely liver abscesses. Spleen leasion is also abscess or a minor infarction. Liver tumefaction most likely is focal nodal hyperplasia, but HCC is also possible. After sanation of the inflammatory leasions, surgical treatment is recommended.
Patohistological biopsy result oft he hypervascular tumor of the left liver lobe:	Microscopic: cylindrical samples of the liver parenchyma-fibrosis. In the parts of the tissue visible lymphocyte infiltration, plasma cells, neutrophilic and eosinophilic granulocytes. Morphological image consistent with cirrhosis, deposit disease or inflammatory myofibroblastic tumor.

Parenteral antimicrobial therapy was continued with metronidazole (24 days), meropenem (17 days), vancomycin (18 days) and fluconazole (21 days). After administering vancomycin he developed a rash and itching. Laboratory parameters revealed CRP 187-21 mg / L, L 14.6-4.11, DBC neutrophils 88.7-35.4%, ly 8.7-41%, eo 0.4-12.6%, mo 1.6-9.51%, ba 0.44-1.54%, T 661, AST 78-31 U / L, ALT 198-41 U / L, GGT 163 U / L, LDH 235-183 U / L, and other findings including red blood cells, kidney parameters, blood glucose, electrolytes, coagulogram, ceruloplasmin, serum copper, acid status, and urine were in referent values.

After radiological abdominal reevaluation in another hospital, a surgeon indicated a needle biopsy of the lesion under ultrasound control but the patient collapsed during a procedure so it was delayed and successfully done after 7 days under CT control. The patient became afebrile with improvement of general condition and regression of the inflammatory markers. After the hospitalisation the infectologist and gastroenterologist recommended to continue with conservative treatment with metronidazole per os 3x500 mg for 4 more weeks.

After some time the patient visited a surgeon at the Clinic for Surgery, Clinical Center Merkur in Zagreb for a consultation. He recommended a operative treatment of the liver tumor after complete regression of the inflammatory liver lesions. The surgical treatment of the liver tumor in the S4 was performed after 5 months without any complications. Today the patient is a healthy and working capable man without any health problems.

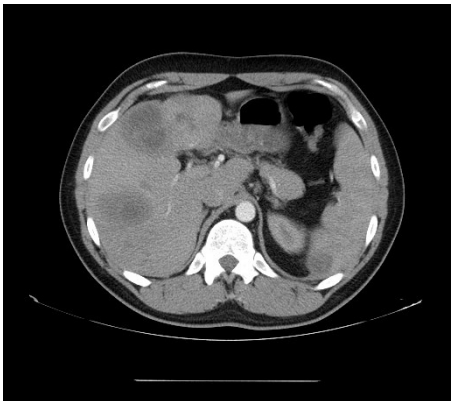

Figure 1. MSCT liver made in the University Hospital of Mostar, Department of Radiology

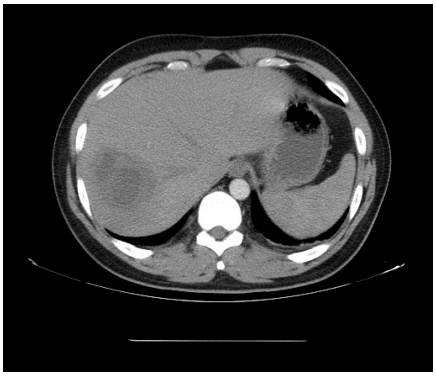

Figure 2. MSCT liver made in the University Hospital of Mostar, Department of Radiology

DISCUSSION

We presented a thirty five years old immunocompetent patient who was admitted to our hospital as a fever of unknown origin with high inflammatory parameters and polymorph nonspecific symptoms. This case report is educational wanting to point out importance and need for a multidisciplinary approach to a complex patient with rare liver diseases. In this process sometimes it is important to aknowledge the very limits of our institution in diagnostic approach and cooperate with other medical centers for the purpose of fast recovery of the patient.

Differential- diagnostic approach to a patient with a fever of unknown origin should consider more often abscesses of parenchymatous abdominal organs as a possible cause. The choice of diagnostic method for the differentiation benign from malignant liver lesions must be rational because these lesions are not easily radiologically differentiated. Focal nodular hyperplasia (FNH) and FNH-like lesions are hypervascular masses that can mimic hepatocellular carcinoma (HCC) and are difficult to differentiate by the same conventional radiological methods [13].

The study of differentiation of benign from malignant liver tumors in children indicates the limitations and advantages of radiological diagnostic procedures in assessing children's liver lesions [14]. Therapeutic approach to symptomatic patients and undefined liver lesions, or high suspicion of focal nodular hyperplasia requires a surgical or intervention approach. Surgery for focal nodular hyperplasia is a safe choice with low morbidity and very good long-term results regarding the quality of life after surgery and is certainly part of a modern therapeutic approach to focal nodular hyperplasia [15-16].

With wide-ranging ultrasound applications, accidental discovery of benign liver tumors often poses a need for a surgical treatment in many young patients. In order to obtain a specific diagnosis and determine the appropriatetreatment,avoidingunnecessaryoperations for asymptomatic tumors with benign evolution or vice versa for surgical treatment of malignant lesions, the surgeon must be aware of the various properties of benign tumors, their expected course, and various radiological techniques for recording positive diagnosis [17]. Computed tomography is essential for the diagnosis and treatment of liver abscesses, and the most important is percutaneous drainage combined with early antimicrobial therapy [18]. Liver abscesses mortality in developing countries is 2-12%, increasing due to an open surgical drainage [8-11].

CONCLUSION

A diagnostic and therapeutic approach to a patient with a fever of unknown origin without leading symptom must always raise a doubt of possible liver abscess, which is rare especially in a young immunocompetent patients, as a cause. Diagnostic and therapeutic approach to a liver lesions is always multidisciplinary and presents a major challenge for every clinician.

SAŽETAK

Uvod: Inflamatorni miofibroblastini tumor (IMT) je rijedak tumor češći kod mlađih ljudi, do 30. godine života, najčešće između 6 i 10 godina. Najčešće je zahvaćen gastrointestinalni trakt i pluća, a može se pojaviti na nekoliko mjesta u isto vrijeme. Klinička slika ovisi o položaju tumora i zahvaćenom sustavu u tijelu pa je vrlo teško postaviti dijagnozu bez kirurške ekstirpacije i patohistološke analize. Odstranjenje u cijelosti u najvećem broju slučajeva znači izlječenje i rijetko se javlja recidiv. Apscesi jetre su češći u žena s rizičnim faktorima dijabetesa, bolesti jetre i rjeđe granulomatoznim bolestima. Mortalitet apscesa jetre u zemljama u razvoju je oko 2-12 %, a povećava se s otvorenom kiruškom drenažom.
Prikaz slučaja: Prikazali smo 35-godišnjeg zdravog bolesnika hospitaliziranog u Klinici za infektivne bolesti Svučilišne bolnice Mostar, KCU Sarajevo Klinika za infektivne bolesti u kolovozu i rujnu 2018. godine. Korišteni su podaci iz medicinske dokumentacije. Imunokompetentni bolesnik hospitaliziran zbog vrućice u trajanju od 8 dana sa zimicom bez tresavice, lošim općim stanjem i visokim upalnim paramterima, a kojem se dijagnostičkom obradom na jetri i slezeni verificiraju multiple fokalne nekrotične promjene koje diferencijalno dijagnostički odgovaraju apscesu i sumnji na malignitet jetre. Etiologija se razjasni nakon iglene biopsije promjena i patohistološke analize u drugom centru te se potvrdi dijagnoza multiplih apscesa jetre u IV i VI segmentu s inflamtornim fibroblastnim tumorom u IV segmentu jetre. Nakon konzervativnog tretmana u trajanju od 8 tjedana apscesi u cijelosti regrediraju, a fibroblastni tumor se kirurški ekstirpira u Kliničkoj bolnici Merkur, Klinika za kirurgiju u Zagrebu u prosincu 2018. godine te tijek liječenja je proveo u potpunosti bez komplikacija.
Zaključak: Pristup bolesniku s multiplim apscesima i tumorom jetre zahtijeva subspecijalizirana iskustva i hitni multidisciplinarni dijagnostičko terapijski pristup istog kako bi se spriječile daljnje komplikacije i smrtni ishod.

Ključne riječi: inflamatorni fibroblastni tumor, apsces jetre, antibiotic

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