Recurrent pain and swelling of the right forearm in a 34 year-old patient

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Case presentation

A 34 year-old female patient with no significant family or medical history, presented for pain, swelling and redness of her right forearm that last for 2 weeks, occurring after strenuous household activities. She reports experiencing similar episodes that resolved spontaneously several times since childhood. At the moment of examination there were no pathological findings at the physical exam (right forearm without pain on palpation, skin discoloration or temperature changes). Laboratory tests were within normal limits.

Musculoskeletal ultrasound performed at the level of the forearm revealed an ill defined mass in the medial 1/3 of the flexor profundus compartment characterized by a mixture of hypo-anechoic and hyperechoic components on longitudinal and transverse gray scale ultrasonography (fig 1). In addition, this mass showed vascularization on color Doppler with flow increasing during compression with transducer.

Subsequently MRI was performed and revealed an intramuscular structure at the level of the deep antero-medial and partially of the posterior compartment of the right forearm, 10/5 cm in size, occupying an important part of the flexor digitoris profundus muscle. MR angiogram showed intense late contrast enhancement of the antero-medial compartment (fig 2).

Questions:
1. What is the diagnosis?
2. Can you provide an explanation for intermittent occurrence of the algic syndrome in this case?
3. Are any other investigations necessary in this case?
4. What treatment options are there?
Cystic tumor of the Liver

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1. What is your diagnosis?
The most likely diagnosis is cyst-adenoma of the liver, since after arterial enhancement, no wash-out is visible in the late phases neither on CEUS, nor in contrast CT. Also the echoic content present in some of the cysts did not enhance following contrast. The differential diagnosis should consider a cyst-adenocarcinoma.

2. Which other tests, if any, do you need for the final diagnosis?
In order to differentiate the two entities an echoguided core biopsy was performed. The conclusion of the pathological exam was: Hyperplasic type alterations of the hepatocytes (regenerative/focal nodular, adenomatous) with no dysplasia.

3. What therapeutic approach would you suggest?
Since cyst-adenomas have a 10% risk of malignant transformation [1] surgical treatment should be recommended, but in the current case, considering the cysts’ dimension, only liver transplantation can be performed.

Liver cyst-adenomas are rare tumors of the liver; they represent less than 5% of the non-hydatid cysts of the liver [2], with less than 200 cases reported worldwide. They originate from the bile ducts in the liver, or less frequently in the extrahepatic bile duct or gallbladder [3,4]. Their etiology is unknown, but is thought to result from ectopic rests of primitive foregut sequestered within the liver or due to the obstruction of a congenitally aberrant bile duct [4]. Environmental and hormonal factors have also been considered [5,6].

The incidence is highest in middle-aged women [7], unlike in our case presentation. Small cysts are asymptomatic, while in larger cysts onset clinical symptoms are not specific, the most frequent ones being abdominal discomfort and abdominal mass, in 1/3 of cases obstructive jaundice being the first symptom to appear [7]. Rarely ascites secondary to portal hypertension or acute abdomen due to intracystic hemorrhage, cyst rupture or torsion, can occur [1,2].

Usually the biological parameters are almost normal, with slightly increased aminotransferases or cholestasis in some cases. Tumor markers have no value in differentiating benign cyst-adenoma from cyst-adenocarcinoma [7].

Ultrasoundography is a useful tool for the first assessment of cyst-adenoma. Usually a well-defined, smooth-walled cyst with septa is seen. On CT the aspect is similar, the cyst being a low-density one. The internal septa and wall enhance following contrast both in CEUS and in contrast CT. It is difficult to distinguish cyst-adenoma from cyst-adenocarcinoma on imaging methods alone. Thick walls, calcification, enhancing papillary projections inside the cyst usually indicate a cyst-adenocarcinoma [8].

Since there is a high risk of malignant transformation, and since patients treated by partial excision, marsupialization, internal drainage, aspiration or intratumoral sclerosing application have experienced high recurrence rates, radical excision of the tumor is the first treatment choice [9]. Ultrasound follow-up every 6 months post surgery should be performed due to the high risk of recurrence. In very large, multiple cysts, such as the case in our patient (in which almost no healthy tissue was available), liver transplantation should be considered.

References


Erratum


For an unexplainable error M. Matucci Cerinic has been included among the authors of the paper. The correct names of authors are: Vlad V, Micu M, Porta F, Radunovic G, Nestorova R, Petranova T, Iagnocco A. The authors request is to remove the M. Matucci Cerinic name from the on-line publication of the paper.