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Case Report

Solitary Fibrous Tumor of Common Bile Duct. Case Report and Literature Review

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Abstract

Solitary fibrous tumor (SFT) is a rare tumor of fibroblastic origin and not fully clear biological behavior. The common bile duct is extremely infrequent location for this neoplasia, being not described in the current literature sources. This report presents a case of SFT in the common bile duct, in 69-year-old male patient and the outcome, as well as literature review of the diagnostic pathological identification of SFT, sites of its origin, and other factors for its behavior, prognosis and treatment.

Keywords: *Solitary Fibrous Tumor (SFT); Bile Duct*

Introduction

Solitary fibrous tumor (SFT) is the rare mesenchymal neoplasia of the fibroblastic origin. This entity was first described by Klemperer and Rabin in 1931 in pleura [1]. SFT equally affects male and female patients; the age varies from 20 up to 70 years old [2]. The majority of SFTs are totally benign, having aggressive features only in 15% of cases [3]. The main prognostic factors of SFT are the clinical stage, tumor's size, mitotic activity [4]. Grossly, tumor presents sharply demarcated nodule, white or grey color, frequently containing cysts and hemorrhages. The size of the lesion varies from 1 up to 20 cm, median- 5 - 10 cm [5]. Histologically, neoplasia presents as the hypercellular or randomly separated fields of spindle, seldom - ovoid, cells with scarce cytoplasm. The fibrous stroma, as well perivascular hyalinosis, is very well developed. The areas of myxoid or edematous pattern are able to occur, mitotic index is low [6].

Despite the leading predominance of SFT, some others sites of SFT are described in modern literature, including nasal cavity and sinuses [7], orbit [8], mediastinum [9], breast [10] and genitourinary system [11]. SFT is also reported to occur in pancreatico-biliary zone; the lesion is described to be located in the liver [12] and 16 cases- in pancreas [13]. Nevertheless, our data base search doesn't provide us any case report of SFT located in common biliary duct (CBD).

Concerning the rarity of this entity- SFT of CBD, we report our case in patient, 69 years old.

Case Report

The Male, white patient, 69 y.o., delivered by ambulance to the Botkin Moscow City hospital in September 2015, clinically with 12-days existing mechanical jaundice (common bilirubin 265 Mmol/l).

At the delivery to the hospital emergency, Abdominal Ultrasound revealed dilatation of the right (4.8 mm) and left (5.2 mm) lobular bile ducts. The round mass, diameter 22 mm, of decreased density is located some more distal to their confluence. The retrograde endoscopic cholangiography with papillotomy and bile ducts revision was first performed in order to release the mechanic jaundice. The

prolonged stricture on the level of the common liver duct 1 (by Bissmutt-Corlette) was found during the operation. Extrahepatic cholangiocarcinoma was suspected and bile-duodenal stenting by means of the plastic graft was made.

CT in multiphase regimen proved the presence of the tumor mass, measured 20 x 21 x 25 mm, in the area of the cystic duct and common hepatic duct confluence. This mass was reported to apply to the portal vein and the right hepatic artery, but without signs of invasion into them. The serum levels of the onco-markers (CA 19-9, CEA, AFP) were normal. After that the patient was comprehensively examined following the general clinical standard protocol. Some associated diseases, including chronic coronary ischemia, essential hypertension, left ureteral- and nephrolithiasis with hydro nephrosis treated by means of nephrostomy, were found during the detailed clinical examination.

Clinical symptoms and signs associated with radiography revealed the diagnosis: hilar cholangiocarcinoma, T2-3NxM0, decompensated stricture of the common bile duct, Bismuth-Corlette I. The patient was surgically operated, the laparotomy, cholecystectomy, trihepatojejunoanastovosis, abdominal cavity drainage was performed as planned. The neoplastic lesion, measured up to 20 mm, in the middle third of the common bile duct, outgoing from its wall, was intraoperationally revealed. Proximal part of the CBD was resected up to the bifurcation following trihepatojejunoanastomosis formation.

Post-operative period was satisfactory, and the patient was discharged home on the 10th day after the surgical procedure.

Pathology revealed grossly, the tumor, 15 mm in the greatest dimension, white to grey color, fibrous monomorphic appearance. Histologically, the lesion consisted of the growth of spindle cells with mild to moderate polymorphism (Figure 1). IHC demonstrated positive expression CD34 (Figure 2), S-100, Bcl-2 (Figure 3), collagen IV type (Figure 4). Desmin, SMA, Dog1, CD117 were negative, index labelling of Ki-67- 3%.

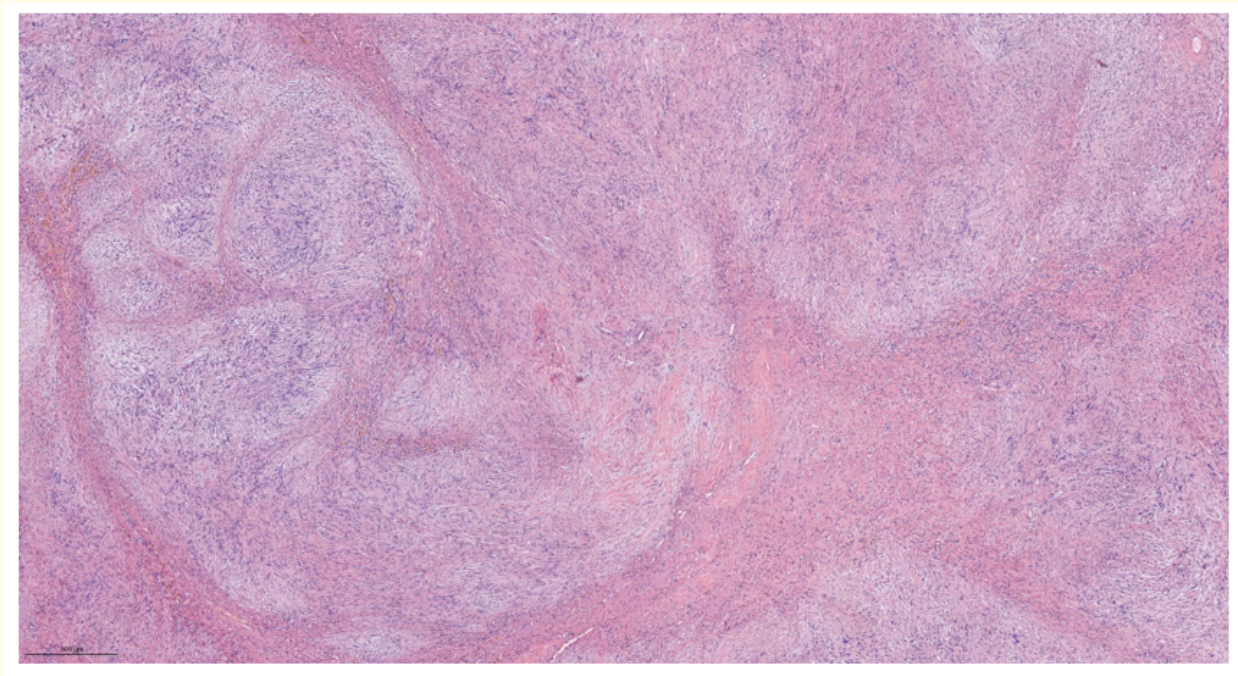

Figure 1

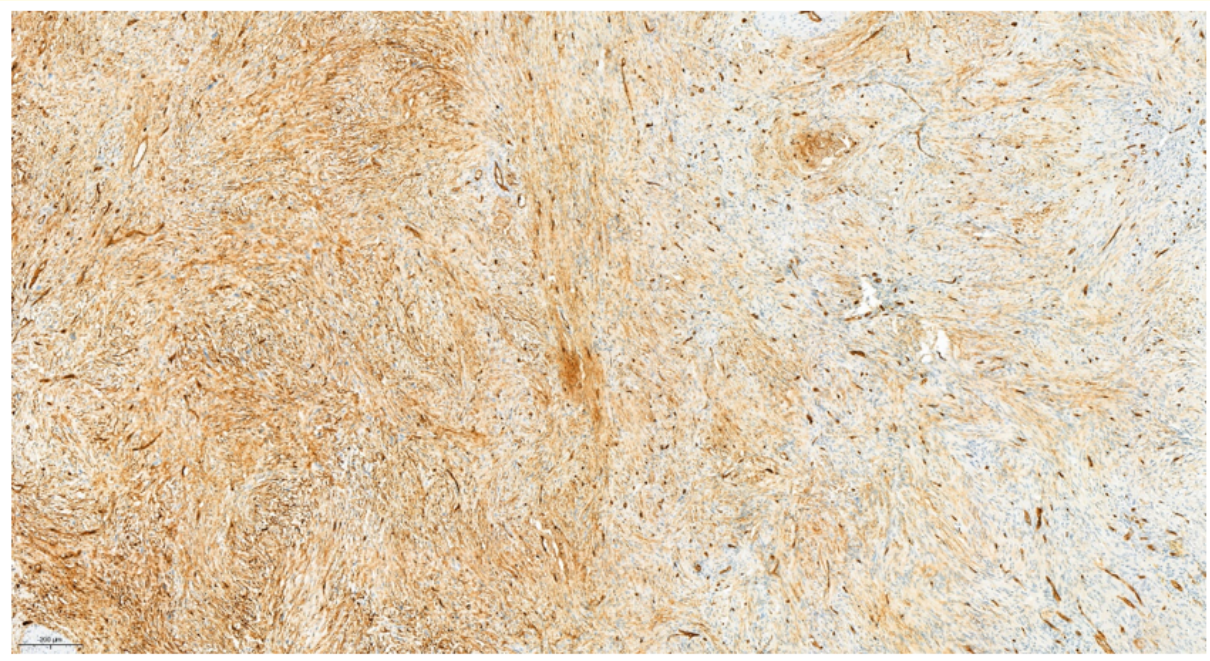

Figure 2

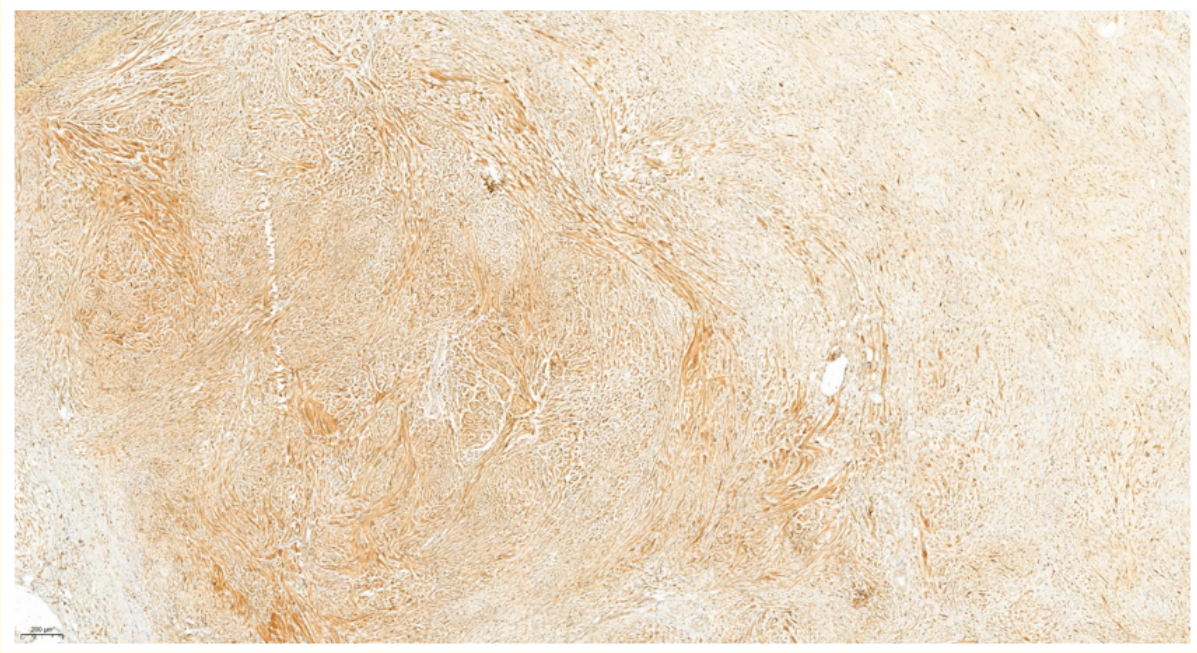

Figure 3

Figure 4

Diagnosis: Solitary fibrous tumor (tumor with undetermined (intermediate) malignant potential) of the common bile duct. ICD-O - 8815/1.

Discussion

We reported the rare case of SFT in CBD. Mesenchymal tumors of CBD are extremely infrequent neoplasia. They are hard to be perioperatively diagnosed, because it lacks either specific clinical signs or radiologic features. Clinically, the patient develops the signs reflecting the affection of the organ. Radiologically, SFT's pattern of enhancement may be moderate or marked, homo- or heterogeneous, tumor also varies in size, shape and margins [6,7]. Differential diagnosis includes such entities as: leiomyosarcoma, liposarcoma, fibrosarcoma, fibrous histiocytoma, hemangiopericytoma, lymphangioma, schwannoma, as well cholangiocarcinoma [12,14,16]. Concerning the treatment, in vast majority the surgery is the only option. If entire tumor can't be removed some doctors suggest radiation therapy. Chemotherapy is rare option and applies particularly in cases of the tumor's spread to adjusted structures and organs [15,16]. The active surveillance for the patient after surgical treatment is strongly recommended, because SFT is the tumor with unknown (intermediate) malignant potential, and in some cases the additional surgery may be required [16]. In conclusion, it must be noted that adjuvant chemotherapy is not indicated in SFT because of the lack of the absence of the sufficient and representative clinical series.

Conclusion

The complex approach to the treatment, including the modern endoscopic decompressive, and, later, radical surgical treatment following by the skilled gross and histological investigation together with IHC, afforded us to state the correct diagnosis, and not to apply unnecessary chemotherapy and radiology. The further patient monitoring revealed no signs of the disease during 28 months follow-up period.

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