Clinical Case Report

Medicine®

OPEN

Ureteral inflammatory myofibroblastic tumor

A case report and literature review

Faping Li, MD, Hui Guo, MD, Heping Qiu, MM, Yuchuan Hou, MD*

Abstract

Rationale: Inflammatory myofibroblastic tumor (IMT) is a rare soft-tissue neoplasm which has been described in a variety of locations. In the urogenital system, IMT predominantly occurs in the bladder and the kidney. IMT arising from the ureter is exceedingly rare and has been sporadically reported before.

Patient concerns: We reported an extremely exceptional case of IMT arising from the ureteral submucosa in a 54-year-old man. The patient was hospitalized with the main complaint of intermittent and moderate left abdominal pain for 2 months.

Diagnoses and Interventions: Computed tomography scan revealed a nearly circular mass in the left upper ureter. Ureteroscopy showed that the ureteral lumen mucosa was smooth. However, the upper ureter was compressed and narrow. Renal dynamic imaging was performed and the measured glomerular filtration rate was 46.98 mL/min (right renal) and 9.77 mL/min (left renal), respectively. A retroperitoneoscopic radical nephroureterectomy was performed. The histopathologic examination revealed that the soft-tissue neoplasm was mainly composed of myofibroblastic spindle cells proliferation with mixed inflammatory infiltrate, containing lymphocytes, neutrophils, and eosinophils. On immunohistochemical staining, the tumor was positive for smooth muscle actin and Ki-67 (<1%+), indicating a confirmed diagnosis of ureteral IMT.

Outcomes: The patient recovered well with no occurrence of complications. At 3-year follow-up, there was no radiologic evidence of tumor recurrence or metastasis and the man was well.

Lessons: Ureteral IMT is extremely rare and often asymptomatic, resulting in delayed diagnosis. Radiologic evidences may be suggestive of the diagnosis of IMT. However, it is necessary to make an accurate diagnosis in terms of histopathologic assessment. Complete lesion excision is the best therapeutic approach with rare recurrences and excellent survival.

Abbreviations: ALK = anaplastic lymphoma kinase, CD = cluster of differentiation, CT = computed tomography, F = female, GFR = glomerular filtration rate, HHV-8 = Human herpesvirus 8, IMT = inflammatory myofibroblastic tumor, IPT = inflammatory pseudotumor, Lt = left, M = male, MRI = magnetic resonance imaging, NR = not recorded, NU = nephroureterectomy, Rt = right, SMA = smooth muscle actin, SR = segmental resection, UR = ureteral reimplantation.

Keywords: diagnosis, histopathology, immunohistochemical, inflammatory myofibroblastic tumor, ureter

1. Introduction

Inflammatory myofibroblastic tumor (IMT) is a rare soft-tissue neoplasm and characterized histologically by a myofibroblastic spindle cell proliferation with inflammatory cell infiltration.[1] According to World Health Organization (WHO) classification, IMT is an intermediate tumor that primarily occurs in children and young adults.[2] IMT has been described in multiple anatomic sites, but typically located in liver and biliary tract (32%), lung (27%) and gastrointestinal tract (10%).[3] In the urogenital system, IMT most frequently occurs in the bladder[4] and the kidney.[5] IMT arising from the ureter is extremely rare with an unknown etiology. To the best of our knowledge, there are only 7 cases in the English literatures over recent decades.[6–12] Herein, we report an additional case of ureteral IMT in a middle aged man and perform a literature review to highlight the most common clinical manifestations, the most useful diagnostic methods, and management of IMT.

2. Case presentation

A 54-year-old man presented to the First Hospital of Jilin University (Changchun, China) with the main complaint of intermittent and moderate left abdominal pain for 2 months. Physical examination revealed percussion tenderness over the left kidney region. Gastroenteric history was not significant. Furthermore, medical and surgical histories were unremarkable as well. In addition to the elevated serum creatinine and blood urea nitrogen (1.67 and 20.90 mg/dL, respectively), laboratory findings were nonspecific. Abdominal computed tomography (CT) demonstrated a nearly circular hypodense mass (2.1 × 2.0 cm) arising from the left upper ureter (Fig. 1A). On contrast-enhanced CT scan, the lesion showed heterogeneous enhancement with relatively well-defined margination and no invasion of adjacent lymph nodes (Fig. 1B). Ureteroscopy reviewed that the

Editor: N/A.

The study protocol was approved by the medical center's ethical committee. Informed written consent was obtained from the patient for publication of this case report and accompanying images.

The authors have no funding and conflicts of interest to disclose.

Department of Urology, The First Hospital of Jilin University, Changchun, China.

* Correspondence: Yuchuan Hou, Department of Urology, The First Hospital of Jilin University, Changchun 130021, China (e-mail: hou63@163.com).

Medicine (2018) 97:46(e13177)

Received: 10 July 2018 / Accepted: 17 October 2018

http://dx.doi.org/10.1097/MD.0000000000013177

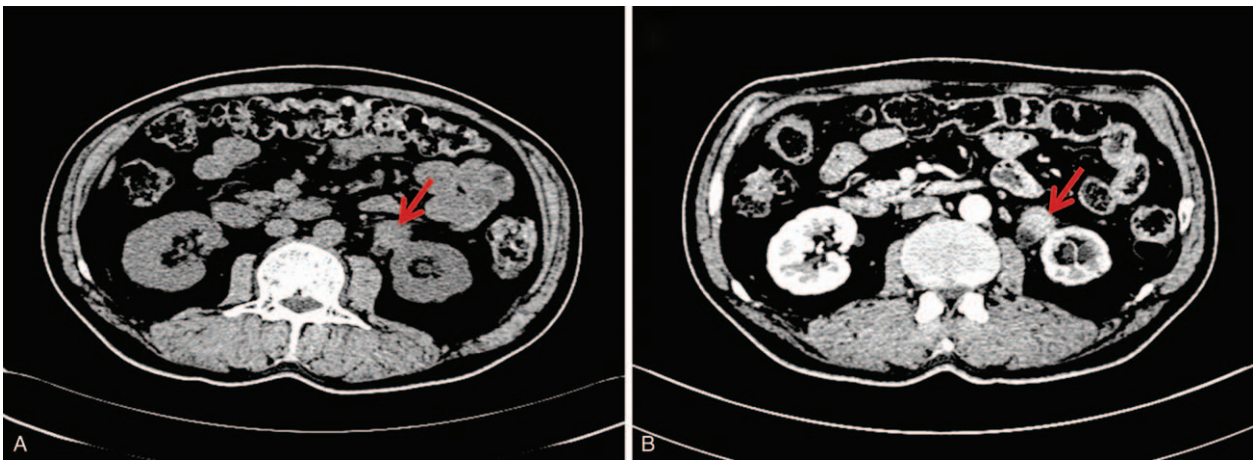

Figure 1. Preoperative contrast-enhanced computed tomography scan: (A) the red arrow indicates a hypodense mass (2.1 × 2.0 cm) arising from the left ureter; (B) the red arrow shows heterogeneous enhancement and relatively well-defined margination.

ureteral lumen mucosa was smooth, while the upper ureter was compressed and narrow. Subsequently, renal dynamic imaging was performed to measure glomerular filtration rate (GFR). The results were 46.98 mL/min of right kidney and 9.77 mL/min of left kidney, respectively.

Due to the left nonfunctional kidney, the patient underwent a retroperitoneoscopic radical nephroureterectomy under general anesthesia. The tumor was located at approximately 3.0 cm beneath the ureteropelvic junction, wrapping the ureter. Because of firmly adhesion with the surrounding tissue, intraoperative blood loss was about 600 mL. The subsequent histopathology revealed a mesenchymal neoplasm mainly composed of myofibroblastic spindle cells proliferation and a mixed inflammatory infiltrate of lymphocytes, neutrophils, and eosinophils (Fig. 2A–C). On immunohistochemical staining, the tumor was positive for smooth muscle actin (SMA) and Ki-67 (<1%+), indicating a confirmed diagnosis of ureteral IMT (Fig. 2D).

The postoperative course was uncomplicated. The patient did not receive any treatment postoperatively and recovered well with no occurrence of complications. At 3-year follow-up, there was no radiologic evidence of tumor recurrence or metastasis and the man was well.

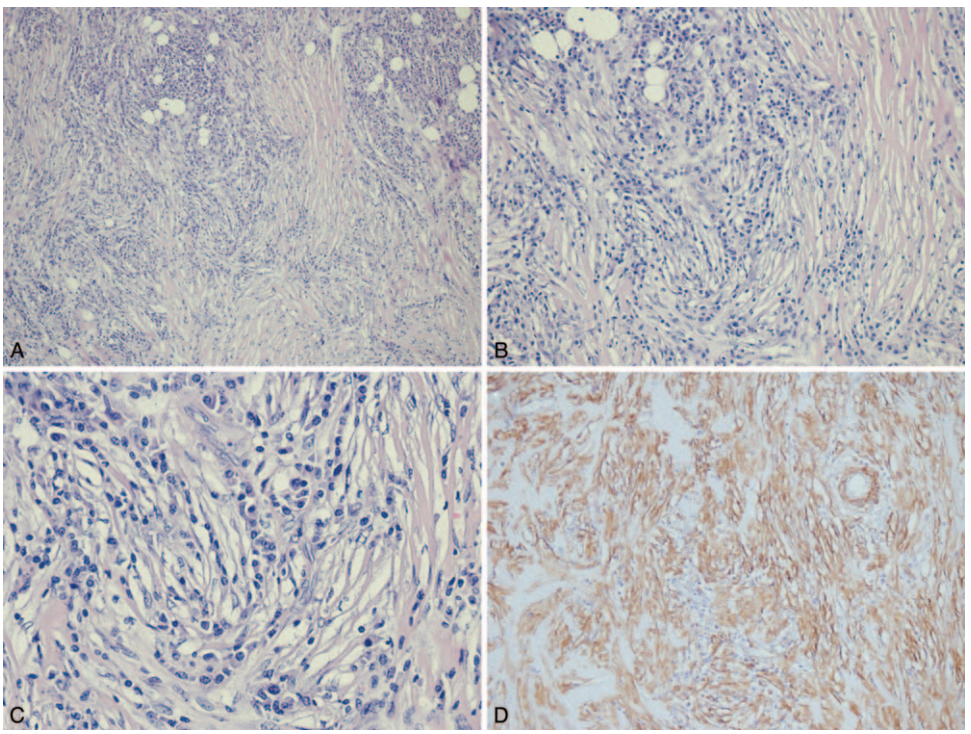

Figure 2. Microscopic examination showed a mesenchymal neoplasm mainly composed of myofibroblastic spindle cells proliferation and a mixed inflammatory infiltrate, containing lymphocytes, neutrophils and eosinophils (A: ×100; B: ×200; C: ×400). Positive for smooth muscle actin expression by immunohistochemical staining (D: ×200).

Table 1

Clinical characteristics of all published cases.

Case report	Age	Sex	Chief complaint	Size, cm	Location in ureter	Treatments	Original diagnosis	Follow-up	Outcome at time of report
[6]	64 y	M	Left renal pain, fever	2.5 × 1.5	Lt lower	NU	IPT	4.1 y	Alive
[7]	54 y	M	Left back pain	4.0 × 3.0	Lt upper	SR	IPT	7 mo	Alive
[8]	8 mo	M	Gross hematuria	4.0 × 3.0	Lt lower	SR + UR	IPT	1 y	Alive
[9]	12 y	M	Hematuria	NR	NR	NU	IMT	NR	Alive
[10]	13 y	M	Left flank pain	1.5 × 1.0	Lt lower	SR	IPT	1 y	Alive
[11]	70 y	M	Gross hematuria	3	Rt upper	NU	IMT	6 mo	Recurrences (1 and 4 mo) Dead (6 mo)
[12]	66 y	M	None	3	Lt upper	NU	IMT	NR	NR
Present case	54 y	M	left abdominal pain	2.1 × 2.0	Lt upper	NU	IMT	2 y	Alive

F = female, IMT = inflammatory myofibroblastic tumor, IPT = inflammatory pseudotumor, Lt = left, M = male, NR = not recorded, NU = nephroureterectomy, Rt = right, SR = segmental resection, UR = ureteral reimplantation.

3. Discussion

The IMT is an extremely rare tumor and predominantly affects children and young adults.[13] The age at presentation varies from 3 to 89 years.[14] IMT has been described in multiple anatomic sites, but typically located in liver and biliary tract (32%), lung (27%), and gastrointestinal tract (10%).[3] In the urogenital system, IMT most frequently occurs in the bladder[4] and the kidney.[5] Nevertheless, IMT arising from the ureter is exceedingly rare. To the best of our knowledge, only 7 cases of IMT arising in the ureter have been published in English since 1997.[6–12] The etiology of IMT remains unclear and the immunologic and infectious hypotheses still need to be validated.[15]

The clinical summary of ureteral IMT is listed in Table 1, including the 7 cases reported in the English literatures and an additional case from the present study. All patients were male. The age of onset ranged from 8 months to 70 years. The mean age of the 8 patients was 41.7 years, and 3 patients were younger than 14 years. Of the 8 patients, there were 4 patients presenting with pain and 3 with hematuria. The size of tumors ranged from 1.5 to 4.0 cm in greatest dimension. The left ureter was more commonly affected. There was no significant difference in the incidence of IMT arising from the upper and lower ureters. In 1 patient, the IMT was accompanied with separate nidus of sarcomatoid carcinoma and high-grade urothelial carcinoma. Unfortunately, the patient died of sarcomatoid carcinoma 6 months after operation. According to the review of the limited literatures, it is premature to establish an epidemiologic profile of ureteral IMT. However, on the basis of the analysis above, we may draw a conclusion that ureteral IMT predominantly affects males with left ureteral predominance and better prognosis.

There are various hypotheses about the pathogenesis of IMT, such as allergic mechanism, immune mechanism, and infection mechanism.[16] However, many proponents believe that the development of IMT is predominantly related to polyfactorial mechanisms including inflammatory response and chromosomal aberrations.[17] Molecular genetics studies have found that anaplastic lymphoma kinase (ALK) gene rearrangement is the initiating factor of IMT formation. A clonal rearrangement of the ALK gene was detected in about 34% to 56% of both pulmonary and extrapulmonary IMT.[18–21] The identification of ALK gene alterations in bladder IMT indicated that they were neoplastic, although the lesions were frequently similar to nodular fasciitis.[22,23] Urinary IMT might be associated with recurrent urinary tract infections and cystoscopy. In addition, it was reported that some drugs such as cyclophosphamide or foreign body in bladder could induce bladder IMT.[24] Moreover, Chen et al reported a case of renal IMT combined with renal calculus, suggesting that IMT might be associated with recurrent and chronic inflammation.[25] In this work, the routine urinalysis of the patient was normal and there was no history of urinary tract infection or urolithiasis. The expression of ALK gene was not detected by immunohistochemical staining.

The clinical manifestations of IMT, which are nonspecific, depend on multiple factors, including tumor size, location, growth rate and individual tolerance. The main symptoms of IMT arising from ureter are pain and hematuria, followed by fever, abdominal mass, nausea, and so on. In the current case, the patient presented to hospital with the main complaint of left abdominal pain for 2 months. The lesion was located at the upper ureter and encapsulated the ureter, resulting in upper ureteral obstruction and hydronephrosis. The laboratory tests showed high level of serum creatinine and blood urea nitrogen, which were consistent with left nonfunctional kidney (GFR = 9.77 mL/min). Of the 7 cases reported previously, 3 patients suffered from pain, 3 patients suffered from hematuria, and 1 patient suffered from fever. In addition, 1 case was asymptomatic and left hydronephrosis was noted during a medical checkup. After further examination, the tumor was detected. From what has been discussed above, it can thus be concluded that pain is the most common symptom (Table 1).

Ureteral IMT is extremely rare and often asymptomatic, resulting in delayed diagnosis. Although imaging and endoscopic examination play an important role in the diagnosis of IMT, histologic assessment is the gold standard.[26,27] CT scan can confirm the relationship between the lesions and ureter, while ureteroscopy is usually performed when there is a suspect of endoluminal lesions. Ureteroscopy is a useful diagnostic and therapeutic tool because it possesses the functions of removing tumor obstruction and providing biopsy samples. If conditions permit, small samples of the lesion can be collected under ureteroscopy for a biopsy.[28] Differential diagnosis includes ureteral carcinoma, idiopathic ureteral inflammation, retroperitoneal tumor, and tumor invasion from ureteral peripheral tissue.

Data collected from the literatures showed that 4/8 patients underwent CT or ureteroscopy, suggesting solid masses arising from the ureter. These lesions were characterized as a single, well-defined, nearly circular mass, and frequently located at the left ureter (Table 2). Only by imaging examination, the mass can be easily misdiagnosed as retroperitoneal tumor or tumor invasion from ureteral peripheral tissue. Therefore, scintigraphy and

Table 2

Histologic and immunohistochemical characteristics of patients.

Case report (reference number)	Diagnostic tools	Histopathology	Immunohistochemical staining
[6]	Uretero-nephroscopy: tumorous lesion. Scintigraphy	Marked cellularity and dense inflammation and infiltration of the periureteral adipose tissue.	Positive for actin, desmin, vimentin
[7]	CT: a heterogeneous 3 cm. mass in the left upper ureter and left hydronephrosis with thin parenchyma	Collagenous fibrous tissue, infiltrating plasma cells, lymphocytes and histiocytes without atypia	Not available data
[8]	Ultrasound, excretory urogram and voiding cystourethrogram	Fibrovascular tissue with extensive necrosis, chronic inflammation and atypical cells	Positive for desmin
[9]	Not available data	Not available data	Negative for ALK and HHV-8
[10]	Excretory urogram: an obstacle of the distal portion of the left ureter with slight ureterohydronephrosis	Inflammatory and myofibroblastic cells	Positive for SMA
[11]	Cystoscopy, retrograde pyelogram, and extensive biopsies	Spindle to stellate cells in myxoid stroma with scattered lymphocytes	Negative for ALK
[12]	MRI and CT: a 3-cm well-circumscribed mass at the left ureteropelvic junction	Marked proliferation of lymphocytes and plasma cells without atypia against background of loose fibrous stroma	Not available data
Present case	CT: a nearly circular mass measuring about 2.1 × 2.0 cm in the left upper ureter	Myofibroblastic spindle cells proliferation and a mixed inflammatory infiltrate, containing lymphocytes, neutrophils, and eosinophils	Positive for SMA and Ki-67 (<1%+)

ALK = anaplastic lymphoma kinase, CT = computed tomography, HHV-8 = Human herpesvirus 8, MRI = magnetic resonance imaging, SMA = smooth muscle actin.

intravenous pyelogram are inevitably performed for further treatment.[6,10]

Radiologic presentation and contrast-enhancement of ureteral IMT were variable and nonspecific. The final diagnosis of IMT mainly depended on histopathology assessment. The histopathology assessment showed that IMT was usually characterized by spindle cell proliferation with inflammatory cell infiltration, including lymphocytes, neutrophils, and eosinophils (Table 2).

Immunohistochemical findings confirmed the definitive diagnosis when mesenchymal cells were positive for vimentin, desmin, SMA, and S100 protein and negative for cluster of differentiation 34 (CD34).[29] Up to 50% IMT cases were positive for ALK in tumor cells.[30] Moreover, positive ALK status was more common in aggressive tumors and associated with a high recurrence rate.[30,31] Positive expression of SMA, desmin, vimentin, and Ki-67 was reported in both previous and present ureteral cases (Table 2).

At present, due to sporadic reports of ureteral IMT, clinicians have limited experience in treatment. Based on the review of the literatures, it was supported that the complete surgical resection of the lesion as an effective treatment for IMT. The negative margin was an important prognostic indicator for local recurrence. A retrospective study was performed by Ong et al in 28 patients with IMT treated by surgical resection. The researchers found that negative surgical margin was associated with 87% reduction in mortality, and was considered as the most important prognostic indicator for local recurrence.[32]

Surgery was not the only treatment. It was reported that radiotherapy was performed for unresectable lesions, nonresponsive IMT, and was used as an adjunctive therapy for IMT with ALK-1 and Ki-67 expression.[33] In addition, anti-inflammatory drugs combined with corticosteroids were also reported to treat IMT successfully.[34,35] However, most ureteral IMT can lead to upper urinary tract obstruction and hydronephrosis, which may result in renal failure. Therefore, surgical resection is an essential therapeutic tool to relieve obstruction and conserve the function of kidney. The most appropriate surgical procedure depends on the localization and size of the tumor, the relationship with the surrounding structures and the surgeons' experience. Among all reported cases, 5 patients underwent ureteronephrectomy and 2 patients underwent segmental resection. In addition, because the stalk of tumor originated from the lumen of the distal ureter, partial cystectomy, and distal ureterectomy with ureteral reimplantation were performed in 1 patient (Table 1).

The IMT is usually considered as a neoplasm with low malignant potential and low local recurrence rate. The recurrence rate is related to large size, abdominopelvic location, and occurring in the elderly.[14] Literatures reported that the recurrence rate ranged from 2% to 25%.[36] A case of IMT with extrapulmonary recurrence 9 years after the primary resection was reported by Morotti et al.[37] On the contrary, Mergan et al evaluated retrospectively 16 children who underwent surgery for IMT. Then, the researchers found that no IMT recurrence occurred later than 2 years after surgery and follow-up was unnecessary and useless after 3 years.[9]

In addition, aggressive IMT invaded bladder or ureter was not encountered. This confirmed the reports that there was no local recurrence or malignant metastasis in the urologic IMT.[5,38] The patient in the present work had been followed up for 2 years without any sign of recurrence. Among all patients reviewed above, only 1 patient had tumor recurrence and even relapsed twice (1 and 4 months, respectively).

Acknowledgment

The authors thank Professor Yuchuan Hou and our colleagues who provide insight and expertise that greatly assist the research.

Author contributions

Investigation and Resources: Hui Guo, Heping Qiu.
Resources: Hui Guo, Heping Qiu.
Writing – original draft: Faping Li.
Writing – review & editing: Yuchuan Hou.

References

[1] Villalobos VM, Camidge DR. Targeting ALK in inflammatory myofibroblastic tumours. Lancet Respir Med 2018;6:405–7.

[2] Telugu RB, Prabhu AJ, Kalappurayil NB, et al. Clinicopathological study of 18 cases of inflammatory myofibroblastic tumors with reference to ALK-1 expression: 5-year experience in a tertiary care center. J Pathol Transl Med 2017;51:255–63.

[3] Hoehne S, Milzsch M, Adams J, et al. Inflammatory pseudotumor (IPT) and inflammatory myofibroblastic tumor (IMT): a representative literature review occasioned by a rare IMT of the transverse colon in a 9-year-old child. Tumori 2015;101:249–56.

[4] Collin M, Charles A, Barker A, et al. Inflammatory myofibroblastic tumour of the bladder in children: a review. J Pediatr Urol 2015;11:239–45.

[5] Kapusta LR, Weiss MA, Ramsay J, et al. Inflammatory myofibroblastic tumors of the kidney - a clinicopathologic and immunohistochemical study of 12 cases. Am J Surg Pathol 2003;27:658–66.

[6] Horn LC, Reuter S, Biesold M. Inflammatory pseudotumor of the ureter and the urinary bladder. Pathol Res Prac 1997;193:607–12.

[7] Nozawa M, Namba Y, Nishimura K, et al. Inflammatory pseudotumor of the ureter. J Urol 1997;157:945–1945.

[8] Weiser AC, Cheng EY. Pseudosarcomatous myofibroblastic tumor of the distal ureter. J Urol 1999;161:1926–7.

[9] Mergan F, Jaubert F, Sauvat F, et al. Inflammatory myofibroblastic tumor in children: clinical review with anaplastic lymphoma kinase, Epstein-Barr virus, and human herpesvirus 8 detection analysis. J Pediatr Surg 2005;40:1581–6.

[10] Harper L, Michel JL, Riviere JP, et al. Inflammatory pseudotumor of the ureter. J Pediatr Surg 2005;40:597–9.

[11] Montgomery EA, Shuster DD, Burkart AL, et al. Inflammatory myofibroblastic tumors of the urinary tract: a clinicopathologic study of 46 cases, including a malignant example inflammatory fibrosarcoma and a subset associated with high-grade urothelial carcinoma. Am J Surg Pathol 2006;30:1502–12.

[12] Hosokawa C, Tsubakimoto M, Inoue Y, et al. Radiologic features of inflammatory pseudotumor of the ureter. AJR Am J Roentgenol 2007;188:W202–5.

[13] Yagnik V, Chadha A, Chaudhari S, et al. Inflammatory myofibroblastic tumor of the urinary bladder. Urol Ann 2010;2:78–9.

[14] Pothadiyil AJ, Bhat S, Paul F, et al. Inflammatory myofibroblastic tumor of the kidney: a rare renal tumor. J Clin Diagn Res 2016;10:ED17–8.

[15] Luo W, Teng P, Ni Y. A rare cardiac inflammatory myofibroblastic tumor involving aortic valve. J Cardiothorac Surg 2017;12:13.

[16] Kasana V, Rajesh S, Chauhan U, et al. Inflammatory myofibroblastic tumor of liver masquerading as focal nodular hyperplasia in a patient with non-cirrhotic portal hypertension and biliary pancreatitis. Indian J Surg Oncol 2016;7:110–4.

[17] Hansen CC, Eisenbach C, Torres C, et al. Maxillary sinus inflammatory myofibroblastic tumors: a review and case report. Case Rep Oncol Med 2015;2015:953857.

[18] Cessna MH, Zhou H, Sanger WG, et al. Expression of ALK1 and p80 in inflammatory myofibroblastic tumor and its mesenchymal mimics: a study of 135 cases. Mod Pathol 2002;15:931–8.

[19] Voelker HU, Kuehn D, Strehl A, et al. Testicular inflammatory myofibroblastic tumor: a known entity at a very rare site. Case Rep Urol 2017;2017:1410843.

[20] Ono A, Murakami H, Serizawa M, et al. Drastic initial response and subsequent response to two ALK inhibitors in a patient with a highly aggressive ALK-rearranged inflammatory myofibroblastic tumor arising in the pleural cavity. Lung Cancer 2016;99:151–4.

[21] Camela F, Gallucci M, di Palmo E, et al. Pulmonary inflammatory myofibroblastic tumor in children: a case report and brief review of literature. Front Pediatr 2018;6:35.

[22] Freeman A, Geddes N, Munson P, et al. Anaplastic lymphoma kinase (ALK 1) staining and molecular analysis in inflammatory myofibroblastic tumours of the bladder: a preliminary clinicopathological study of nine cases and review of the literature. Mod Pathol 2004;17:765–71.

[23] Tsuzuki T, Magi-Galluzzi C, Epstein JI. ALK-1 expression in inflammatory myofibroblastic tumor of the urinary bladder. Am J Surg Pathol 2004;28:1609–14.

[24] Razi A, Radmehr A. Inflammatory pseudotumor of bladder: report of 2 cases and review of literature. Urol J 2008;5:62–5. 2008.

[25] CY C, JH L, YM L. Inflammatory myofibroblastic tumor of the kidney associated with a renal stone (Chinese). JTUA 2008;19:235–8.

[26] Lamas-Pinheiro R, Rodesch G, Devalck C, et al. Pulmonary myofibroblastic tumour involving the pericardium and left atrium in an 18 month infant. Ann Thorac Cardiovas 2016;22:312–4.

[27] Ma ZH, Tian XF, Ma J, et al. Inflammatory pseudotumor of the spleen: a case report and review of published cases. Oncol Lett 2013;5:1955–7.

[28] Vashistha V, Shabsigh A, Zynger DL. Utility and diagnostic accuracy of ureteroscopic biopsy in upper tract urothelial carcinoma. Arch Pathol Lab Med 2013;137:400–7.

[29] Appak YC, Sahin GE, Ayhan S, et al. Inflammatory myofibroblastic tumor of the colon with an unusual presentation of intestinal intussusception. European J Pediatr Surg Rep 2014;2:54–7.

[30] Coffin CM, Hornick JL, Fletcher CDM. Inflammatory myofibroblastic tumor - comparison of clinicopathologic, histologic, and immunohistochemical features including ALK expression in atypical and aggressive cases. Am J Surg Pathol 2007;31:509–20.

[31] Alaggio R, Cecchetto G, Bisogno G, et al. Inflammatory myofibroblastic tumors in childhood: a report from the Italian Cooperative Group studies. Cancer 2010;116:216–26.

[32] Ong HS, Ji T, Zhang CP, et al. Head and neck inflammatory myofibroblastic tumor (IMT): evaluation of clinicopathologic and prognostic features. Oral Oncol 2012;48:141–8.

[33] AlKindi MG. A rare case of inflammatory myofibroblastic tumor of the mandible mimicking a malignant tumor. Saudi Dent J 2017; 29:36–40.

[34] Segawa Y, Yasumatsu R, Shiratsuchi H, et al. Inflammatory pseudotumor in head and neck. Auris Nasus Larynx 2014;41:321–4.

[35] Berger A, Kim C, Hagstrom N, et al. Successful preoperative treatment of pediatric bladder inflammatory myofibroblastic tumor with anti-inflammatory therapy. Urol 2007;70:372.e13-372.e15.

[36] Li J, Yin W-h, Takeuchi K, et al. Inflammatory myofibroblastic tumor with RANBP2 and ALK gene rearrangement: a report of two cases and literature review. Diagn Pathol 2013;8:147.

[37] Morotti RA, Legman MD, Kerkar N, et al. Pediatric inflammatory myofibroblastic tumor with late metastasis to the lung: case report and review of the literature. Pediatr Dev Pathol 2005;8:224–9.

[38] Karnak I, Senocak ME, Ciftci AO, et al. Inflammatory myofibroblastic tumor in children: diagnosis and treatment. J Pediatr Surg 2001; 36:908–12.