Une hernie inguinale chez un homme à utérus

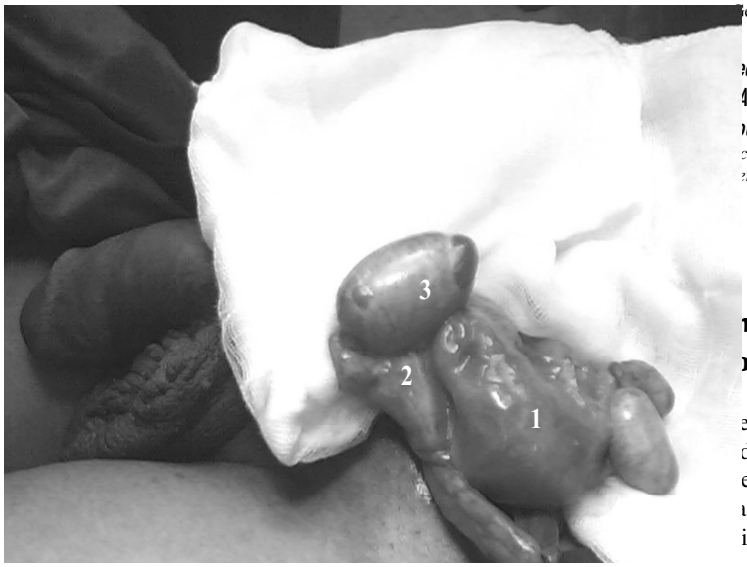
L'homme à utérus, nommé aussi syndrome de persistance des canaux de Müller (SPCM), correspond à la présence d'un utérus et deux trompes chez un individu normalement virilisé. Il s'agit d'une malformation anatomique assez rare, due dans 85 % des cas, à une anomalie au niveau d'une glycoprotéine secrétée par les cellules de Sertoli : l'hormone anti müllérienne (AMH) [1, 2]. Deux anomalies génétiques en sont responsables : un défaut d'expression du gène AMH (19 p13,3) [3] ou une mutation affectant le gène du récepteur de type II de l'AMH (12q13) [4]. Nous rapportons une nouvelle observation de SPCM.

Observation

Nous rapportons le cas de Mr. Ali âgé de 26 ans, sans antécédents pathologiques particuliers, qui a été hospitalisé pour cure chirurgicale d'une hernie inguinale gauche. A l'interrogatoire, Mr. Ali n'avait pas de facteurs d'hyperpression intra abdominale. A l'examen, il présentait une hernie inguino-scrotale gauche non compliquée. Par ailleurs, l'examen du scrotum ne trouvait pas de testicule droit.

En per opératoire, la dissection de la région inguinale gauche, découvre un gros sac de hernie oblique externe gauche. Le cordon spermatique gauche jusque là n'a pas été identifié. A l'ouverture du sac, on constate une formation tissulaire ferme reliée par deux pertuis à deux structures ovalaires qui ont le même aspect mais de taille différente ; l'une de ces deux structures (2 x 3 cm) était retirée du scrotum ; l'autre plus petite (1 x 2 cm) était au niveau de l'orifice inguinal profond (Figure 1). L'ensemble faisait évoquer un utérus avec deux trompes et deux gonades ressemblant plutôt à des testicules.

Figure 1 : Après ouverture du sac herniaire, on découvre un utérus (1), deux trompes (2) et deux gonades: une grosse (3) qui était retirée de la bourse et une petite (4) retirée de l'orifice inguinal profond.



Une biopsie de ces deux gonades a été réalisée. La dissection a permis de réaliser une déconnection du sac herniaire de ces différentes structures. Après fermeture de son collet, le sac herniaire a été réséqué. La structure rappelant l'utérus n'a pas été réséquée, du fait que l'un des deux canaux déférent se perd dans l'épaisseur de cette structure. La gonade la plus volumineuse a été replacée dans la bourse gauche ; la petite avait un pédicule très court, elle a été abandonnée avec « l'utérus » dans la région inguinale gauche. L'intervention a été complétée par une cure pariétale à la Bassini. Les suites opératoires immédiates ont été simples, le malade a été mis sortant au deuxième jour post opératoire.

L'examen histologique des biopsies a confirmé que les 2 gonades étaient bien des testicules. Un caryotype a été réalisé, il était normal (46 XY). Deux spermogrammes, réalisés à 1mois et à 2 ans de l'intervention, avaient conclu à une oligo-asthéno-spermie.

Conclusion

L'existence d'une hernie inguinale et d'une cryptorchidie controlatérale doit faire évoquer le SPCM. La découverte d'un utérus et des trompes chez un homme au cours d'une intervention chirurgicale pour hernie inguinale, doit faire rechercher les canaux déférents qui doivent être soigneusement préservés. Une hystérectomie est à proscrire si on risque de faire léser ou fragiliser les canaux déférents. Méconnaître cette anomalie anatomique, risque de compromettre une fertilité déjà précaire.

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ncreatic tuberculosis mimicking pancreatic oplasia

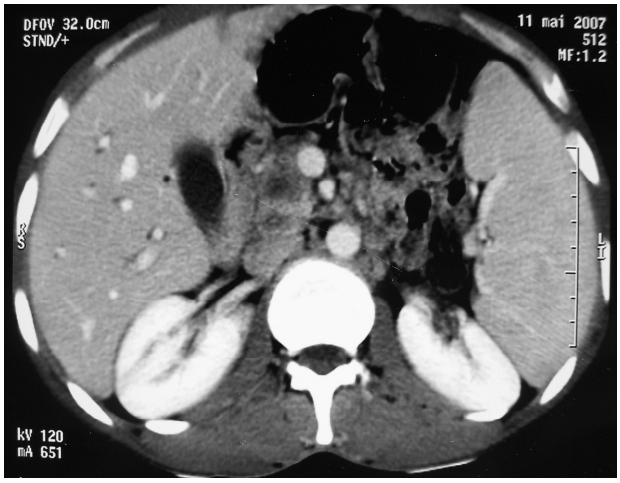
erculosis involving the pancreas or the pancreatic bed and draining peripancreatic nodes is rare [1, 2]. Clinical entation is protean, posing diagnostic challenges. We report ase of pancreatic tuberculosis which masqueraded as a inoma of the head of the pancreas.

Case report

A 32-year-old man was admitted with a two-month history of mild cholestatic jaundice, epigastric pain associated with weakness. He had severe anorexia and 8 kilogram weight loss. Other family members had no health problems. On physical examination, he had mild icterus and abdominal tenderness with deep palpation. The liver was not enlarged and the gallbladder was not palpable. In blood analysis, hemogram showed no abnormalities, bilirubin was 37 mmol/l (reference range : 17 mmol/L), conjugated fraction 6.0 mmol/L (reference range 7 mmol/L), alkaline phosphatase was 260 IU/L (reference range : 30-120IU/L), aspartate amino transferases and alanine amino transferases were 153 IU/L (reference range: 0-35 IU/L) and 88 IU/L (reference range: 0-35 IU/L) respectively. Serum amylase was 73 U/L (reference range: 60-180 U/L). Serology for HIV was negative.

In abdominal ultrasonography intra and extra-hepatic bile ducts were dilated. A computed tomography (CT) scan confirmed the presence of a 3 x 3 cm solid mass in the head of the pancreas with central necrosis and peripancreatic lymphadenopathy (figure 1).

Figure 1 : Hypodense necrotic lesion in the head of the pancreas.



CA 19-9 rate were elevated (295UI/l). A preliminary diagnosis of a periampullary was made. Laparotomy was performed and during the surgical procedure a large firm mass involving the head of the pancreas and the hepatoduodenal and hepatogastric ligaments was identified with adenopathy in the surrounding area. Intraoperative frozen section histological examination of multiple nodal biopsies showed granulomas with giant cells and no evidence of malignancy. Cholecystectomy and bilio-duodenal anastomosis were performed. More lymph nodes and the mass biopsies were obtained for specific histological examination. This confirmed the extended necrosis, with caseous granulomatous inflammation of possible tuberculous etiology: Ziehl-Neelsen staining for acid-fast bacilli was positive. Culture of the tissue was found positive for Mycobacterium tuberculosis (figure 2). Specific medical treatment with isoniazid, pyrazinamide and rifampin was started (pyrazinamide was suspended after 2 months). Six months later, the patient have no symptoms neither jaundice; the CT control shows a partial resolution of the pancreatic mass (figure 3).

Figure 2 : Peripancreatic lymph node. Broad foci of caseating necrosis

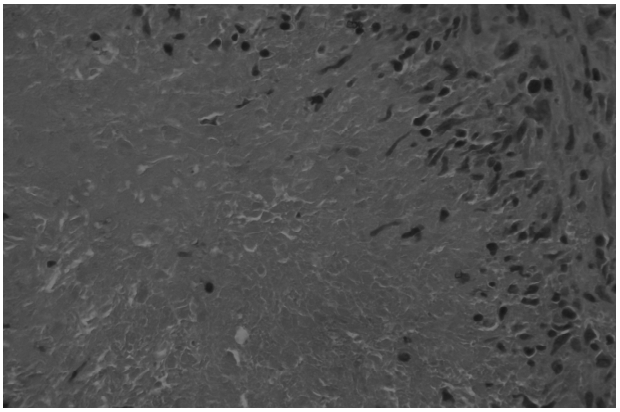
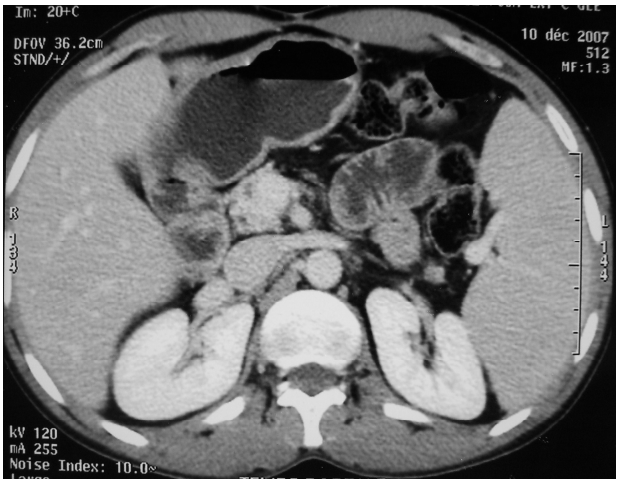


Figure 3 : CT control after a six-month anti-tuberculosis therapy: partial resolution of the mass.



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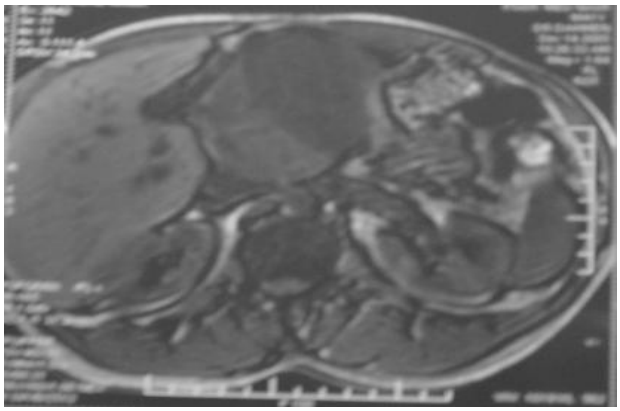
Retroperitoneal myofibroblastic inflammatory tumor

Inflammatory myofibroblastic tumor, also known as inflammatory pseudotumor, is an uncommon tumor, characterized by a controversial etiology, various histopathologic features and an unpredictable biological behavior. They can be found virtually at any anatomic site, with a predilection for the lung, the genito-urinary tract and the mesentery [1]. Retroperitoneal location has been rarely reported.

Case report

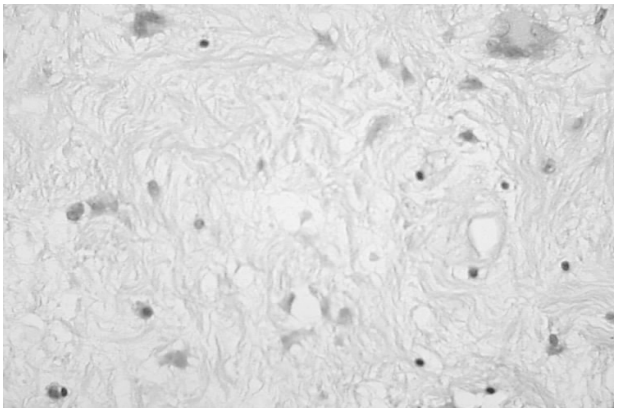
A 41-year-old man presented with history of epigastric pain, abdominal lump of 2 years duration. Physical examination revealed a deeply located mass at abdominal palpation. There were no evident foci of infection. The leukocyte count was normal. A hypochromic microcytic anemia was found. The platelet count was normal. Computerized tomography and magnetic resonance imaging revealed a large mass with well-defined borders, located between the stomach small curvature, pancreas and the liver (Fig. 1), without calcification or contrast enhancement, raising the suspicion of a mesenchymal tumor. A laparotomy was performed. The mass was retro-gastric, adherent to the pancreas and the portal veins. The tumor was partially resected. No palliative treatment was administrated. Post-operatively, the patient presented infectious complications and died 3 months later after the initial diagnosis.

Figure 1 : Magnetic resonance imaging: Large retroperitoneal mass compressing the liver



Histologically, the tumor was well circumscribed, of moderate cellularity, composed of bland spindle or plump cells dispersed in a loose-textured and myxoid stroma (Fig. 2). The cytoplasm was eosionophilic or amphophilic without cross-striations. The nuclei were round or elongated, with dispersed chromatin and inconspicuous nucleoli. Multinucleate histiocytoid cells with an eosinophilic or vacuolated cytoplasm were also seen (Fig. 2). The tumor cells were intermingled with an inflammatory infiltrate consisting of plasmocytes, small lymphocytes and occasional eosinophils. Nuclear atypia, mitoses and foci of necrosis were not identified.

Figure 2 : Hematoxylin-eosin x200: Bland fusiform and multinucleate cells admixed with inflammatory cells, mainly lymphocytes within a myxoid stroma.



Immunohistochemically, the tumor cells were consistent with a myofibroblastic phenotype; they expressed strongly vimentin (1:100; Dako, Glostrup, Denmark) and smooth muscle actin (1:100; Dako) (Fig. 3). Cytokeratin (1:75; Dako) and PS100 (1:50; Dako) were negative. There was no expression of anaplastic lymphoma kinase ALK (1:50; Dako) protein. A marked overexpression of protein p53 (1:25; Dako) by tumor cells was found (Fig. 4). Special stains for mycobacteria and fungi were negative. In situ hybridization (PNA probes, Dako) performed to detect Epstein-Barr virus (EBV) was negative. Investigation of Human Herpesvirus-8 (HHV8) by polymerase chain reaction (PCR) was also negative. The histological diagnosis was of an inflammatory myofibroblastic tumor of the retro peritoneum.

Figure 3 : Immunohistochemistry x200: The tumor cells express smooth muscle actin

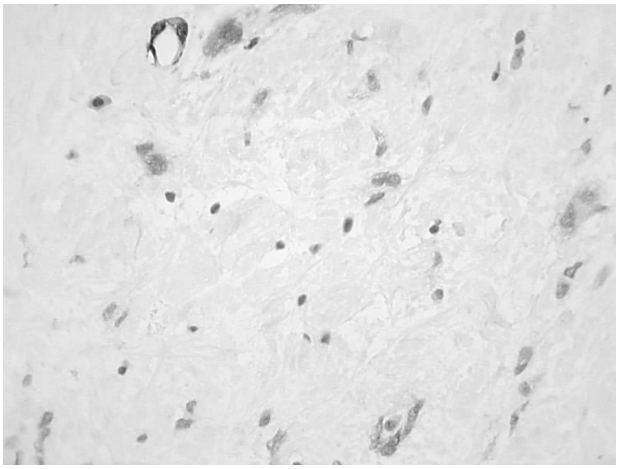
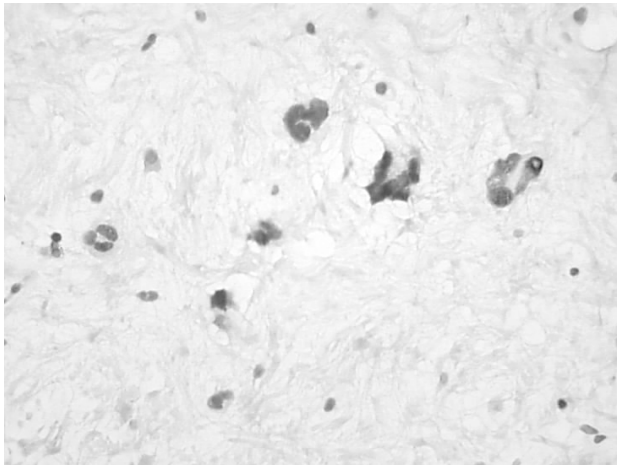


Figure 4 : Immunohistochemistry x200: Nuclear expression of p53 by tumor cells



Conclusion

Inflammatory myofibroblastic tumors are rare lesions of still controversial etiology with an uncertain biological potential that can range from a frequently benign course to a more aggressive evolution.

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Le dermatofibrosarcome de Darier-Ferrand de l'enfant dans une forme angiomateuse

Le dermatofibrosarcome de Darier- Ferrand (DFSDF) est une tumeur mésenchymateuse cutanée rare, de malignité intermédiaire, caractérisée par une évolution lente, une extension locale, un faible pouvoir métastatique mais une forte tendance à la récurrence. Le DFSDF est encore plus rare chez l'enfant (1, 2). Il représente 5 à 6% des tumeurs de l'enfant. Au début, le diagnostic reste souvent longtemps méconnu en raison de la lenteur d'évolution ainsi que de l'aspect clinique souvent trompeur ressemblant à une chéloïde, une morphée ou un angiome.

Nous rapportons deux observations de DFSDF dans leur forme angiomateuse.

Observation 1

S.R., jeune fille de 18 ans, sans antécédent pathologique particulier avait présenté depuis l'âge de 3 ans, un nodule de la région inter mammaire pour lequel elle n'a jamais consulté. A l'âge de 18ans, ce nodule ressemblant à une chéloïde, a subi de façon inappropriée des infiltrations de corticoïdes retard, ce qui a entraîné une augmentation importante de la taille de la lésion en 3 mois, avec tendance au saignement important. L'examen cutané pratiqué à l'âge de 18 ans, avait montré une tumeur angiomateuse de 12 cm de grand axe, mamelonnée, infiltrée, saignant au contact. Les aires ganglionnaires étaient libres. Le reste de l'examen somatique était normal. L'échographie doppler avait montré un flux artériel pouvant cadrer avec une malformation artério- veineuse. L'IRM a écarté la nature vasculaire de cette tumeur. La radiographie de thorax ainsi que l'échographie abdomino- pelvienne étaient normales. La malade avait été traitée chirurgicalement à deux reprises par une exérèse simple de la tumeur puis par une exérèse plus large (4 cm de marge) (figure 1), devant la confirmation histologique et immuno-histochimique du diagnostic de DFSDF. L'examen histologique avait révélé une prolifération intradermique faite de cellules fusiformes disposées en travées irrégulières parallèles prenant un aspect tourbillonnant (figure 2). Le stroma était très riche en vaisseaux sanguins. L'examen immuno-histochimique avait montré une positivité avec les anticorps CD34. L'évolution était maquée par l'absence de récurrence après un recul de trois ans.

Figure 1 : Tumeur excisée

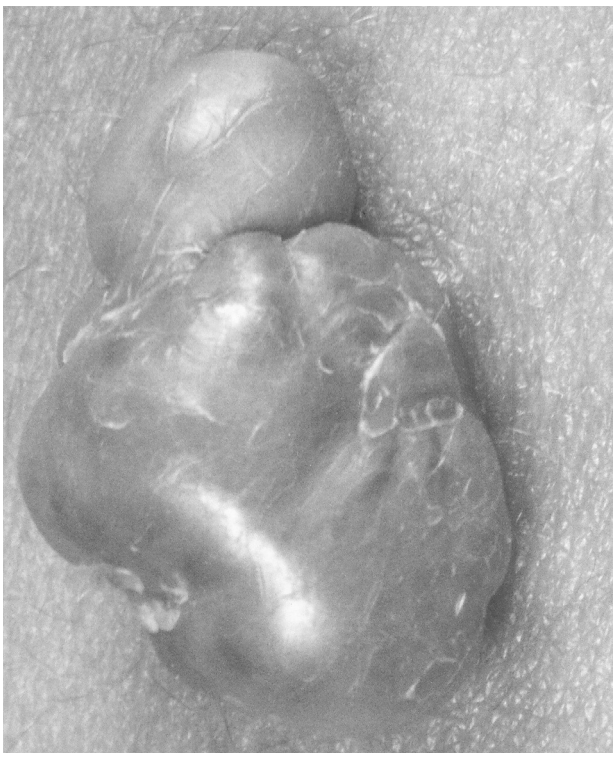
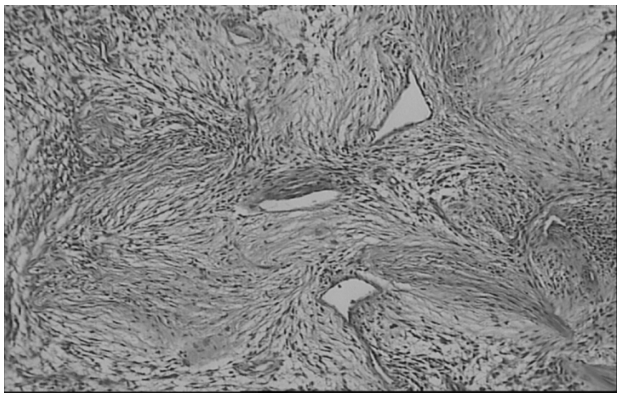


Figure 2 : Prolifération dermique de cellules fusiformes prenant un aspect tourbillonnant



Observation 2

Il s'agissait d'une jeune fille de 36 ans, sans antécédent pathologique particulier, qui présentait depuis la naissance, une tumeur angiomateuse de 3 cm de diamètre du dos, ayant doublé de taille depuis 6 mois, sans facteur déclenchant notable. L'examen cutané avait trouvé une tumeur angiomateuse bilobée de 7 cm de grand axe, non pulsatile, non douloureuse (figure 3). Les aires ganglionnaires étaient libres. Le reste de l'examen somatique était sans particularité. L'échographie couplée au doppler a montré une tumeur vascularisée. L'IRM a éliminé le diagnostic d'une malformation artério-veineuse et la malade a été confiée aux chirurgiens pour une exérèse chirurgicale large (marges de 4 cm) devant la suspicion de DFSDF. L'examen histologique de la pièce d'exérèse avait confirmé ce diagnostic en montrant une prolifération dermique dense de cellules fusiformes de type fibroblastique disposées de façon storiforme avec de rares mitoses et atypies cytonucléaires. Le stroma était riche en vaisseaux sanguins et en fibres de réticuline. L'immuno-histochimie avait révélé une positivité du CD34. L'évolution était marquée par l'absence de récurrences après 1 an d'évolution (figure 4).

Figure 3 : Aspect avant exérèse

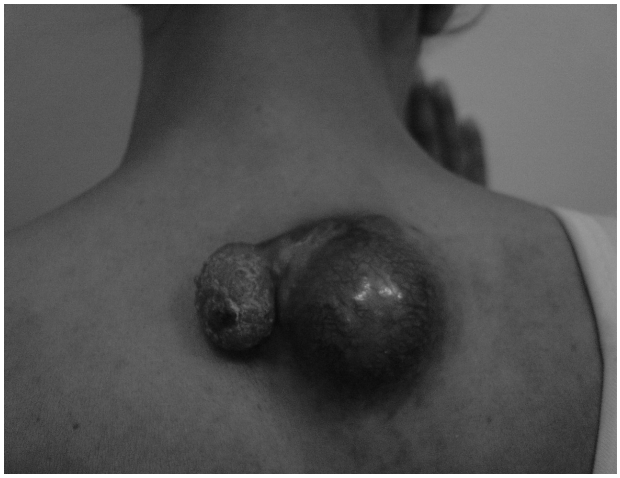
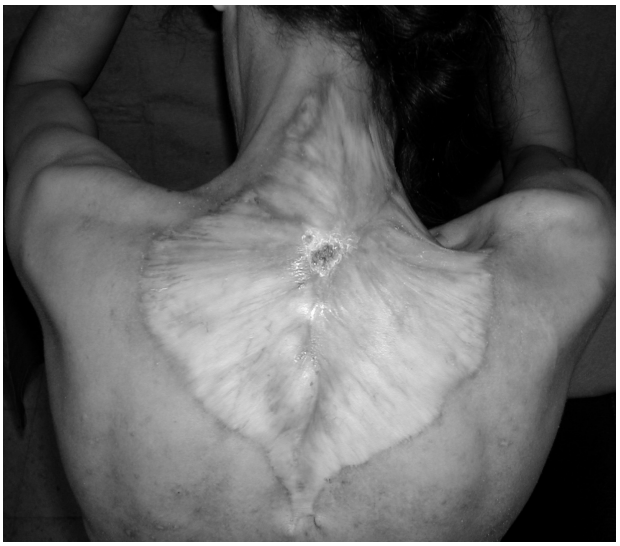


Figure 4 : Aspect après exérèse



Conclusion

Devant la difficulté diagnostique du DFSDF chez l'enfant, la prise en charge doit être multidisciplinaire.

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Une lithiase vésicale sur un ballonnet de sonde vésicale

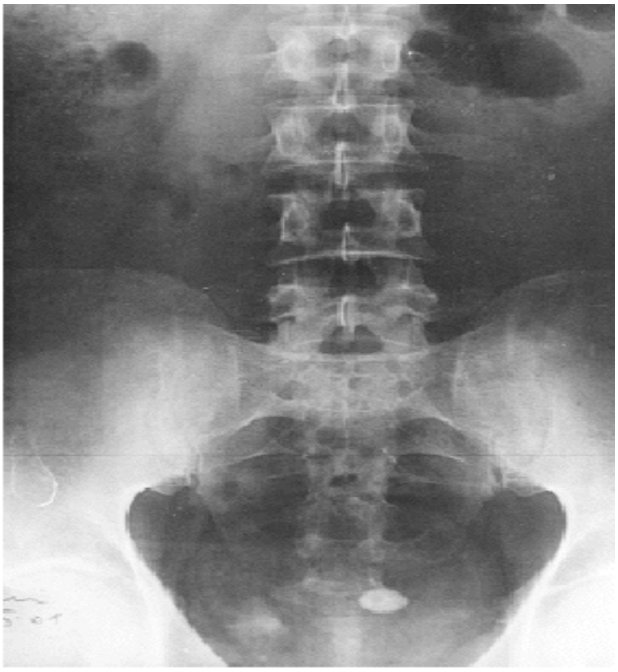
Les corps étrangers de la vessie peuvent être de nature et d'étiologie très diverses (1-3). Nous rapportons le cas d'une lithiase vésicale développée au dépens d'un fragment de ballonnet de sonde vésicale type Foley.

Observation

Melle BN, âgée de 31 ans, appendicectomisée il y a 5 ans, a consulté en urgence pour des douleurs pelviennes évoluant par intermittence associées à une hématurie terminale, des impériosités mictionnelles et des brûlures mictionnelles exacerbées en fin de miction. L'examen clinique était sans

particularité en dehors d'une sensibilité hypogastrique. L'abdomen urinaire sans préparation (AUSP) avait objectivé une opacité de tonalité calcique se projetant sur l'aire vésicale, de 2 cm de grand axe avec un centre peu opaque (Figure 1). L'examen échographique confirme le diagnostic de lithiase vésicale et montre un aspect normal du haut appareil urinaire.

Figure 1 : AUSP : Opacité de tonalité calcique se projetant sur l'aire pelvienne avec un centre peu opaque.

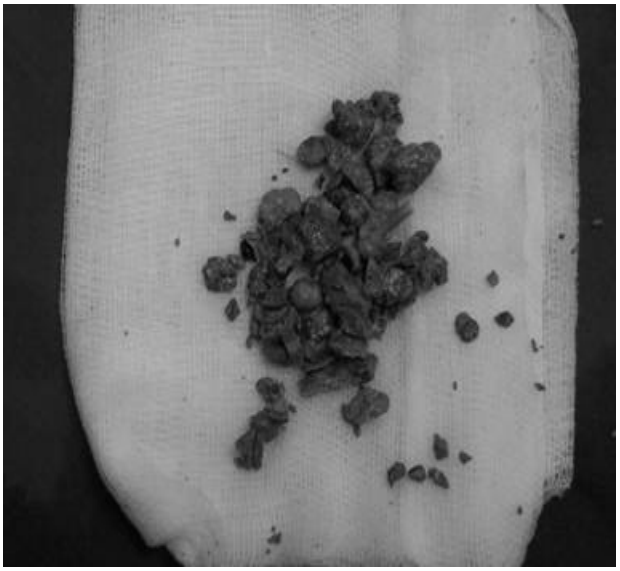


L'examen cytobactériologique des urines n'a pas isolé de germe. Sous rachianesthésie, la patiente a eu une cystoscopie confirmant le diagnostic. La lithiase a été traitée par lithotripsie balistique endo-corporelle. Lors de la fragmentation du calcul un nucleus mou s'est apparu et il s'avère être un fragment de ballonnet d'une sonde vésicale type Foley (Figure 2). En reprenant l'anamnèse, une sonde vésicale a été mise en place lors de l'intervention quelle a eu il y a 5 ans avec une chute spontanée de celle-ci en postopératoire. Le diagnostic d'une lithiase vésicale développée sur un corps étranger a été retenu. Il s'agissait d'un fragment du ballonnet de la sonde vésicale laissée en place suite à son explosion par hyper-remplissage.

Conclusion

Les lithiases vésicales sur corps étrangers ne sont pas rares. Il faut y penser devant les antécédents du patient et l'apparition des troubles mictionnels inexpliqués. L'AUSP et l'échographie font le diagnostic. La méthode d'extraction dépend de leur taille et de leur mobilité, elle fait en général appel aux techniques endoscopiques ou mini invasives. Dans tous les cas, la prévention passe par la vérification de l'intégrité du matériel ce qui permet de reconnaître la présence de ce corps étranger et de procéder précocement à son extraction.

Figure 2 : Lithiase vésicale fragmentée centrée par un fragment de ballonnet de sonde type Foley.



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Asymptomatic chronic torsion of a pelvic wandering spleen

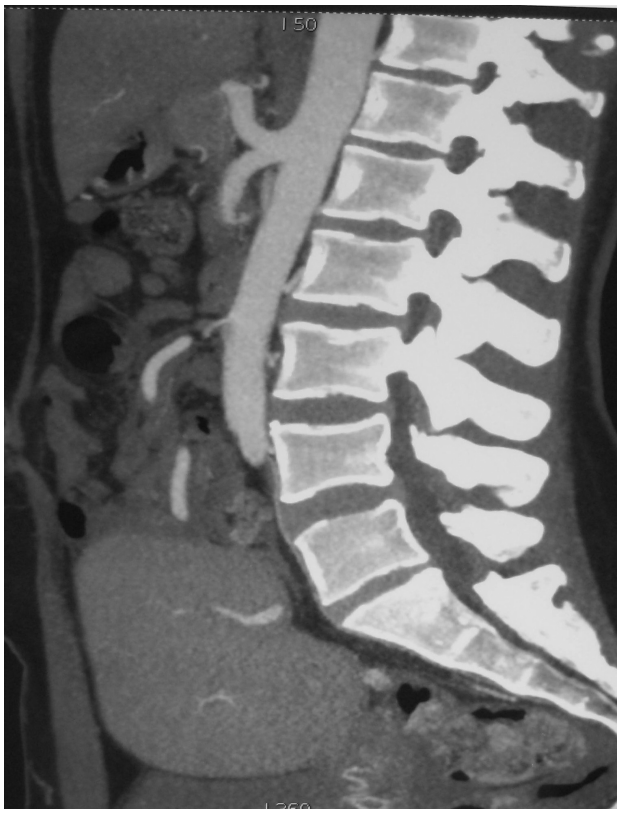
Wandering spleen also known as ectopic, free-floating, splenoptosis or aberrant spleen is characterized by migration of the spleen from the left upper quadrant to a more caudal location in the abdomen. It may be congenital due to absence or underdevelopment of gastrosplenic and splenorenal ligaments or acquired due to weakened supporting splenic ligaments (1, 2). These two forms of anomalies result in the formation of a long vascular pedicle that makes the spleen hypermobile, predisposing it to torsion. Wandering spleen is quite rare anomaly with a reported incidence of less than 0.5% in several large series of splenectomies (3). Torsion of the pedicle is what makes symptoms appear that may be acute, chronic or

intermittent. We describe herein an asymptomatic chronic torsion of a wandering spleen in a 29 year-old woman.

Case report

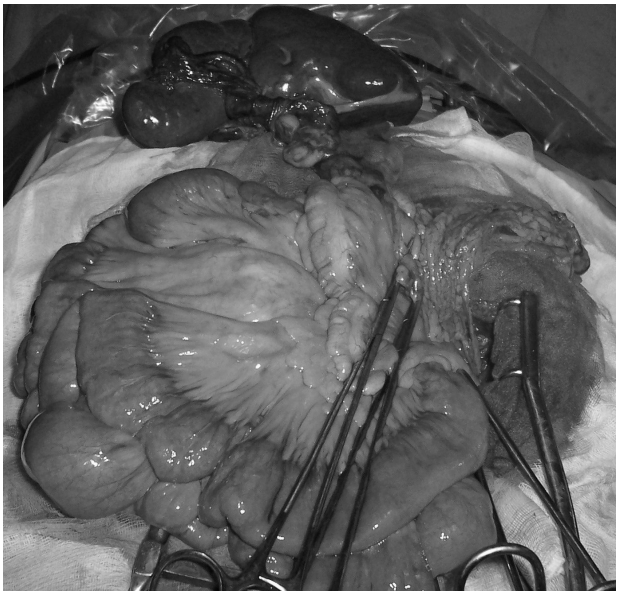
A 29 year-old woman presented with an asymptomatic and mobile pelvic mass. On further questioning, she gave a history of an asymptomatic abdominal mass since the last 2 years which was felt at varying locations in the abdomen. Physical examination revealed a well-defined 9x 8 cm, nontender, reniform, in the left iliac fossa that was mobile and firm in consistency. The rest of the physical examination was unremarkable. Laboratory data showed a reduced platelet count of 93000/mm3, a white blood cell count of 4500/mm3, and a hemoglobin level of 10 g/dl. Ultrasonography revealed the absence of the spleen in the left upper quadrant and a large mass, approximately 20 cm in length, occupying the pelvis, with the features of an ectopic spleen. The mass could be manipulated between the two iliac fossae. We have attempted to do a doppler but we didn't succeed. Axial computed tomography (CT) also showed the absence of the spleen in its normal anatomical position and a spleen-like mass in the pelvis, suggestive of a wandering spleen. A sagital contrast-enhanced CT image showed the enlarged spleen, measuring 19x10 cm, suspended by elongated, dilated, and somewhat tortuous splenic vessels (Figure 1).

Figure 1 : Sagital contrast-enhanced CT image showing a large wandering spleen in the pelvis suspended by an elongated and tortuous vascular pedicle.



In addition CT also revealed multiple collaterals in the hilum of the spleen. There was normal parenchymal enhancement without focal lesions following injection of contrast medium. Upper gastrointestinal endoscopy was normal. The patient was prepared and scheduled for laparoscopic mesh splenopexy with a possibility of splenectomy. At laparoscopy, the left upper quadrant was filled with bowel loops and a bigger spleen with infractions in many sides was found in the right iliac fossa with a long pedicle which had twisted 5 clockwise rotations (Figure 2). Therefore, the decision of splenectomy was taken and because of the enlarged shape of the spleen, it was converted into a laparotomy through a midline incision. The patient made a good postoperative recovery and was discharged after 6 days. Cell blood count shows an increase of platelets count to 250000/mm3 and a hemoglobin rate at 11,5g/dl. Vaccination for encapsulated organisms was performed 2 weeks after splenectomy.

Figure 2 : A bigger spleen with infractions in many sides with a long twisted pedicle



Conclusion

Wandering spleen is a rare clinical finding. This diagnosis should be considered whenever there are mobile abdominal or pelvic mass, signs and symptoms of an acute abdomen or during investigations of chronic intermittent abdominal pain. The vulnerability of this condition predisposes it to various complications. So, surgery is the only definitive treatment for wandering spleen; the choice between splenectomy and splenopexy depending on pre- and intra operative findings of a viable spleen.

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Bartter syndrome revealed at adult age by recurrent nephrolithiasis, associated with hypertension and metabolic syndrome

Bartter syndrome (BS) is a rare genetic salt-losing tubulopathy, first described in 1962, characterized by hypokalemia, hypochloremia, metabolic alkalosis, hyper-reninemia, increased aldosterone secretion and normal blood pressure [1]. Additional features included vasopressin-resistant, impaired urinary concentrating ability and a relative resistance to the pressor effect of angiotensin II, increased urinary prostaglandin excretion was also reported later in this syndrome [2]. Bartter like syndromes are now classified on the basis of the subjacent genetic defect in three major types: distal convoluted tubule dysfunction leading to hypokalemia currently known as Gitelman or Bartter syndrome, the more severe condition of polyuric loop dysfunction referred to as antenatal Bartter or hyperprostaglandin E syndrome, and the most severe condition of combined loop and distal convoluted tubule dysfunction leading to hyperprostaglandin E syndrome with sensori-neural deafness [3]

Patients with BS have a normal or low blood pressure although increased aldosterone. Thus, Bartter like syndromes are considered as a good human model to gain insight into the mechanisms responsible for controlling vascular tone. Despite the frequent occurrence of hypercalciuria and nephrocalcinosis, nephrolithiasis is rarely reported [4, 5].

Here we report a case of BS revealed at 32 years by recurrent and multiple urinary lithiasis spontaneously eliminated. Systematic biologic exams discovered hypokalemia. A 24 hours blood pressure monitoring revealed hypertension. Complementary investigations of hypokalemic hypertension found a metabolic syndrome associated to BS.

Case report

A 32-year-old man was referred to our endocrinology unit by urologist for exploration of hypercalciuria. The patient consulted for spontaneous urethral emission of multiple stones since one year. He reported polyuria and polydipsia since childhood. His parents were third degree consanguineous. He was born by vaginal delivery, his mother had unexplained polyhydramnios during pregnancy. He went to school for 6 years, was working as waiter in a café. He had a family history charged of obesity, hypertension. His father died at 70 years of terminal renal failure of unknown etiology. He was taking no medication nor licorice, nor alcohol. He was 162 cm tall, 78 kg weighted so a body mass index 29.7 kg/m2. His mother and father height were 170 and 180cm respectively, his expected height was 181,5 cm. His blood pressure was 130/70 mmHg, his waist/hip circumference 103 cm/105 cm. He had a triangular visage with small chin. There were no purple stretch marks, bruises, cutaneous capillary fragility, facio-troncular obesity nor facial flushing. Auscultation detected no abdominal bruit, there was no edema. Diuresis was quantified to 6 liters per day. Macroscopic aspect of stones issued by the patient was calcic. Abdominal radiographs showed multiple pyelic and caliciel bilateral radioapacities (Figure 1). Abdominal ultrasound showed two non symmetric small kidneys 85 and 90cm, with decreased differentiation, there was a diffuse hepatic steatosis. Biochemical parameters are summarized in table 1. There was a normocalcemic hypercalciuria with elevated intact parathyroid hormone, hypokalemia with increased urinary potassium, moderate chronic renal failure with 83 ml/min creatinin clearance. A 24 hours blood pressure monitoring revealed hypertension (Figure 2). Water deprivation test showed nephrogenic diabetes insipidus. Further investigations for hypokalemic hypertension were negative, making hypercorticism, pheochromocytoma and 11, hydroxylase deficiency unlikely. There was a hyperaldosteronism with hyper reninemia (Table 2). Doppler ultrasound of renal artery was normal. Parathyroid scan with Tc-99m MIBI injection, bone mineral density were normal.

Table 1 : The patient's biochemical data

	Blood (normal values)	**24-h urines (normal values)**
Creatinine/CC, μmol/L ml/min	125 (62-115)	81.4 (97-137)
Calcium, mmol/L mmol/day	2.20 (2.10-2.55	10.62 (2.5-7.5)
Phosphate, mmol/L mmol/day	1.30 (0.87-1.45)	43 (13-42)
Alkalin phosphatases, IU/L	106 (100-290)	
Potassium, mmol/L mmol/day	2.4 (3.5-5.1)	96 (25-125)
Sodium, mmol/L mmol/day	142 (136-146)	106 (40-220)
Protides, g/L g/day	68 (64-83)	1.1 (0.05-0.15)
Magnesium, mmol/L mmol/day	0.55 (0.65-1.05)	5.65 (3-5)
pH	7.45 (7.35-7.45)	
Bicarbonate, mmol/L	30 (18-23)	
Uric acid, mmol/L	0.46 (0.1-0.35)	
Glucose, g/L	0.83 (0.74-1.1)	
Total cholesterol, g/L	2.3 (<2)	
HDL cholesterol, g/L	0.37 (>0.4)	
Triglycerides, g/L	4.07 (<1.5)	

Figure 1 : Abdominal radiograph, multiple pyelic and caliciel bilateral radioapacities

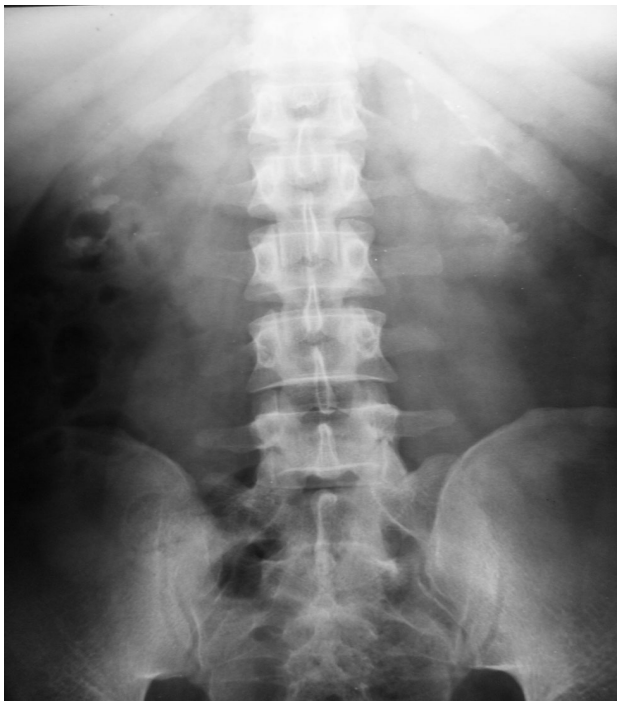
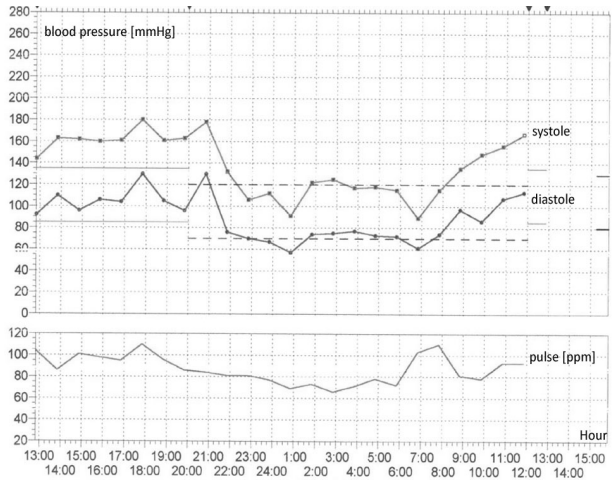


Figure 2 : 24 hours blood pressure monitoring



In view of polyhydramnios, hypokalemia, hypomagnesemia, increased renal potassium and magnesium wasting, metabolic alkalosis, hypercalciuria, hyperreninemic hyperaldosteronism, altered urine concentration, short stature, dysmorphic features, the diagnosis of BS was made. Abdominal obesity, hypertension, hypertriglyceridemia, decreased HDL-cholesterol, defined the metabolic syndrome in our patient.

The patient was prescribed chloride potassium 600 mg/day and spironolactone 50 mg/day then 75 mg/day. Average systolic and diastolic blood pressure improved three months after diagnosis, calciuria decreased slightly but the patient continued calculi emission, renal function decreased gradually, kalemia increased but didn't reach normal values. Ophthalmic exam showed signs of stage II hypertensive retinopathy one year after diagnosis which regressed after, parathormone levels normalized. Two years after diagnosis, creatinin level was 160 μmol/L, creatinin clearance 63.6 ml/min, kalemia 3.8 mmol/L, calcemia 2,44 mmol/L and calciuria 6 mmol/day

Conclusion

Our patient presented with two features uncommonly associated with BS, hypertension and nephrolithiasis. The patient was certainly hypotensive and volume contracted during infancy, and then developed metabolic syndrome which origin is multifactoriel: familial background, insulin-resistance favoured by chronic hypokalemia and hyperaldosteronism

Table 2 : The patient's hormonal exploration

	Blood	24-h urines	Normal values
Parathyroid hormone	129		10-65 (pg/mL)
Prolactin	13.4		2.6-13.1 (ng/mL)
IGF1	290		100-300 (pg/mL)
IGFBP3	4440		1 5 0 0 - 4 3 0 0
Calcitonin	6		(pg/mL)
Basal aldosterone	420		<10 (pg/mL)
2h orthostatism	676		10-180 (pg/mL)
4h orthostatism	1018		30-360
Basal rennin	135		2.8-39.9 (mUI/L)
2h orthostatism	210.4		4.4-46.1
8h basal cortisol	129		87-224 (μg/L)
20h basal cortisol	117		<100
1mgDXM overnight test	11.8		<20
Free cortisol		78	39-348 (μg/24h)
ACTH	5		<50 (pg/mL)
VMA		9	2-10 (mg/day)
Metanephrins bloc		1.9	<2 (mg/day)
Basal 17OH Progestérone	1.4		<5 (ng/mL)
60min ACTH stimulation	1.8		<10

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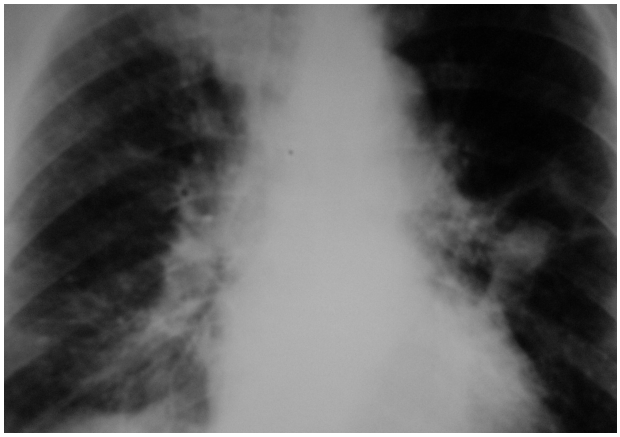
Coexistence of active tuberculosis, cancer and aspergilloma of the lung.

Lung cancer and tuberculosis are two pathologies relatively frequent in our country. Their incidence is at 20 for one hundred thousand inhabitants for the former and 13 for one hundred thousand inhabitants for the latter. Their association and the association of each of them with aspergilloma were previously reported. However, aspergilloma occurs almost always as a complication of tubercular sequellae.
To our knowledge, this is the first case report of active tuberculosis, lung cancer and aspergilloma occurring in the same patient.

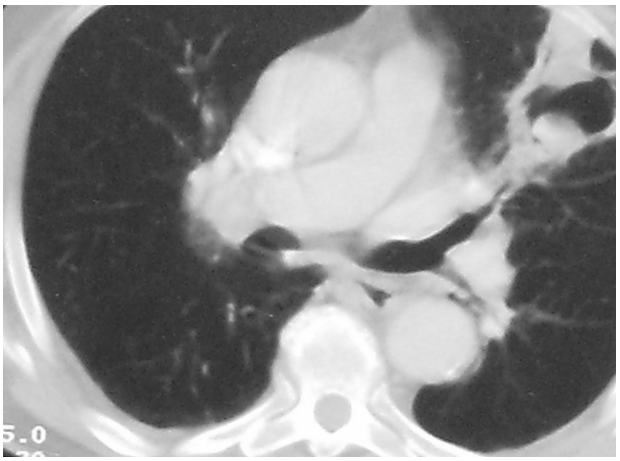
Case report
Mr DM, a 73-year-old man, with a history of 40 -pack-year smoking, non alcoholic, is admitted to our hospital for a massive haemoptysis. He presented 2 years ago a myelodysplasia treated by repeated transfusions.
On physical examination, pallor was noticed. No other abnormalities were present. Sputum samples were tested negative for acid fast bacilli (AFB) by Ziehl Neelsen's staining technique. Conventional method of culture on Lowenstein Jensen's medium did not yield growth of M. tuberculosis. A cavitary lesion in the left upper lobe was found on chest radiograph (Fig.1).

Figure 1 : Chest radiography: cavitary lesion in the left upper lobe.



The lesion showed the “air crescent sign,” characteristic of aspergilloma. A CT scan of the chest revealed a cavity with an intracavitary mass in the left upper lobe (Fig.2). Fiber-optic bronchoscopy was normal. Abdominal ultrasonography revealed splenomegaly, hepatomegaly; and intra peritoneal and retroperitoneal adenopathies. Blood cell analysis showed: haemoglobin: 6.4g/dl, white blood cells: 880.000/mm3, platelets: 127.000/mm3, erythrocyte sedimentation rate: 140 mm/h, total protein level: 90g/l, albumin: 33.5g/l (normal: 59-69g/l), alpha 1 globulin: 2.7g/l (normal: 2-4g/l), alpha 2 globulin: 9.2g/l (normal: 6-11g/l), beta globulin: 5.7 g/l (normal: 8-14g/l), gamma globulin: 38.9g/l (normal: 11-18 g/l), Ig A: 5.55 g/l (normal: 0.8- 3.1g/l), Ig M: 4.08g/l (normal: 0.55-3 g/l), Ig G: 30.97 g/l (normal: 6.5- 15g/l) . Serology of aspergillus was positive.

Figure 2 : Chest CT scan: air crescent sign in the left upper lobe



Consequently, in view of hemoptysis and presence of an aspergilloma surgery was performed (haemoglobin was 8.3g/dl in spite of red cell transfusion). Per-operative exploration revealed a mass measuring 8 cm in diameter located between the culmen and the lingua. As the etiology of this mass was unknown, a resection of the left upper lobe was performed. Gross pathologic examination of the resected specimen showed a cavity in the center of the lobe. On microscopic examination, there was inflammation, bronchiectasis and tuberculoid lesions. The center of the cavity was occuped by a necrotic septate fungal elements of Aspergillus species surrounded by an epidermoid carcinoma. Lymph nodes presented a follicular tuberculoid reaction.
After surgery, the patient had no more haemoptysis but anaemia persisted and transfusions were needed. Antituberculosis treatment was administered the third day post-operatively. It consisted on isoniazid: 3 mg/Kg/day, rifampicin: 10mg/kg/day, ethambutol: 20 mg/kg/day and pyrazinamid: 25 mg/kg/day.
The patient follow –up was 2 months and then he was lost to follow-up. He was asymptomatic, his chest X ray film was normal regardless to an elevated left diaphragm and the AFB were absent of the sputum smear.

Conclusion
The uniqueness of our case report is that the association between lung cancer, active pulmonary tuberculosis and aspergilloma has -to the best of our knowledge- not been reported in the literature before. Many particularities are revealed between the association of the three pathologies in the same patient compared to their associations two by two.

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