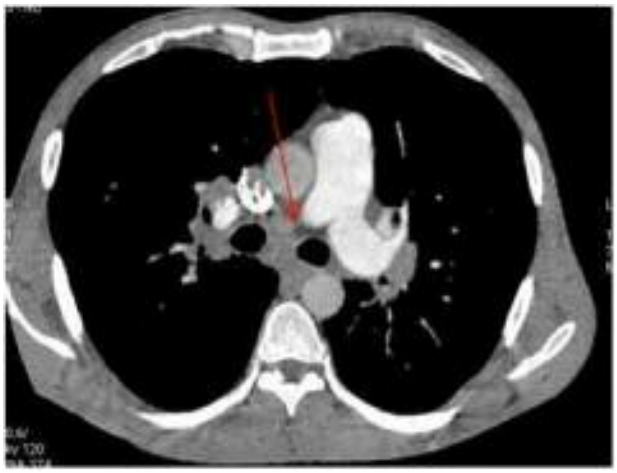
Figure 3 : An axial section of a CT scan of the chest showing thrombosis of the right pulmonary artery.



Conclusion

Antiphospholipid syndrome (APLS) is usually primary, but can be associated with a wide range of other conditions such as systemic lupus erythematosus, Sjögren's syndrome, mixed connective tissue disease (MCTD), idiopathic inflammatory myopathies, infectious diseases, malignancies, and drug-induced conditions. However, the association between APLS and pulmonary LCH has not been described in the literature.

The pathology of pulmonary LCH is poorly understood, but granulomas rich in LCs, eosinophils, macrophages and lymphocytes develop in and destroy distal bronchioles as a result of a cell-mediated immune response. APLS also has an immunological basis related to antiphospholipid antibodies and/or phospholipid binding proteins. In addition, APLS and LCH are multisystem diseases which may affect multiple organs.

Thus, the questions to be answered are whether it is a coincidental association or the two diseases are secondary to similar pathogenic mechanisms and if this association worsens the prognosis of LCH.

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Small cell carcinoma of the ovary of the hypercalcemia type In children

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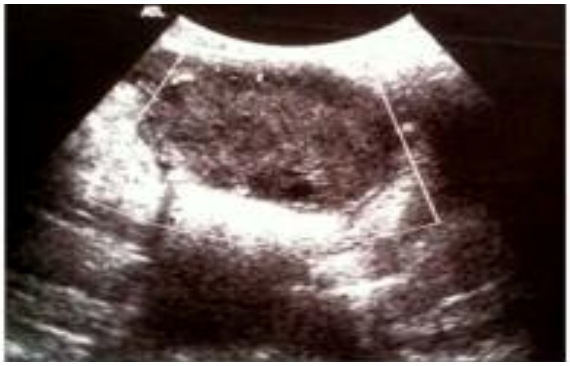
Small cell carcinoma of the ovary of the hypercalcemia type (SCCOHT) is uncommon and aggressive ovarian tumor that primarily affects young women and rarely observed in premenstrual adolescents. Histologically, the typical pattern is diffuse follicle-like sheets of small, closely packed cells with scant cytoplasm. As in epithelial ovarian carcinomas, CA-125 could be a useful marker (1-3). Hypercalcaemia is present at the time of diagnosis in more than 60% of cases with SCCOHT (2-4). We report a case of this unusual tumor whose diagnosis had not been suspected before surgery and we point out its evolution and its management.

Case report

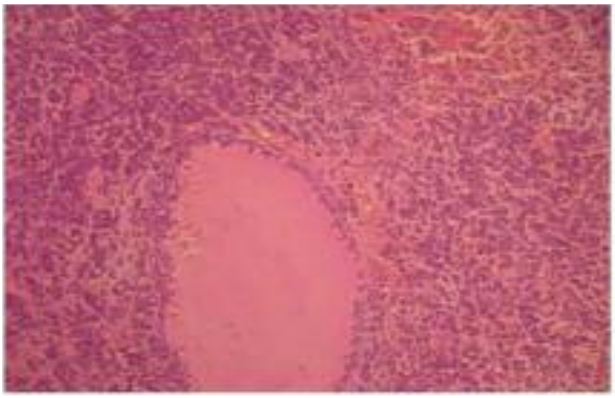
A 10-year-old girl was admitted to our department for acute abdominal pain. She had had an intermittent and isolated pelvic pain for 6 months. A history of weight loss could not be confirmed. On examination, the pelvis was tender without fever or any palpable mass. No features of precocious puberty were noted. Ultrasound examination showed a 6-cm solid right ovarian mass and ascites (figure 1).

Figure 1 : Ultrasound examination showed a 6-cm solid right ovarian mass.



An emergency laparotomy was performed revealing a necrotic right ovary due to torsion of an ovarian solid tumor. The left ovary looked normal and neither ascites nor adhesion was detected in the abdominal cavity. A right salpingo-oophorectomy was therefore performed. Histological and immunophenotypical studies concluded that it was a small-cell carcinoma of the ovary of the hypercalcemic type (figure 2). Subsequent laboratory tests revealed hypercalcemia (2,8 mmol/l). HCG, -FP and serum carcinoembryonic antigen (CA) 125 were normal. Chest x-ray, radionuclide bone scan and cerebral CT scan were normal. The postoperative course was uneventful. In view of the highly malignant form of the tumor, the patient was given 6 courses of chemotherapy based on vinblastine, bleomycin and cisplatin. Seven years after the initial surgery, the patient was free of recurrent disease (normal calcium level, negative staging including chest radiography and abdominal ultrasound).

Figure 2 : Ovarian small cell carcinoma, hypercalcemic type. Prominent follicle-like spaces filled with eosinophilic fluid are present. Tumor cells are round and have scant cytoplasm.



Conclusion

The combination of hypercalcemia and an ovarian mass in premenarchial girls should awaken the suspicion of SCCOHT. SCCOHT had a malignant potential tumor with poor prognosis and complete surgical resection is strongly recommended, even in case of unsuspected diagnosis before surgery. Aggressive chemotherapy after surgery seems to be mandatory.

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Nephrogenic adenoma of the bladder associated with urinary tuberculosis

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The nephrogenic adenoma (NA) constitutes an uncommon benign metaplasia of the urothelial mucosa (1). It is probably underestimated in literature. NA can occur anywhere in the urinary tract, but the most frequent localization was the bladder (3). Our clinical case represents the 9th (1,2,4) report of a NA within an urinary bladder with history of urinary tuberculosis.

The aetiology of NA remains unclear; however, it has been linked to chronic irritating factors, such as trauma, chronic infection, urological surgery, urinary stones, irradiation, urinary catheterization or repeated instrumentation for diagnostic or therapeutic purposes and chemical agents, such as Bacille Calmette-Guerin (3-5). The clinical and the endoscopic appearance of NA are not specific. It can be clinically as well as morphologically mistaken for malignancy of the bladder (5).

Thus, the definite diagnosis is anatomopathological. It is characterized by the presence of ducts lined with epithelium combined with the absence of atypia and mitotic activity. Immunohistochemical staining for nuclear transcription factor for renal development (PAX2) is useful to rule out others diagnosis such as prostatic adenocarcinoma, benign urothelium and urothelial carcinoma (3).

There have been no reports of NA malignant transformation or metastatic lesion (4,5).

NA is mainly treated by transurethral electroresection and administration of antibiotics in case of urinary tract infections (5). In spite of the benign nature of this condition, patients must be closely monitored (urine cytology, ultrasound and cystoscopy) because the rate of recurrence of NA is high (4,5).

Herein, we report a new case of NA in the bladder with history of urinary tuberculosis.

This case will be used to discuss diagnostic criteria of NA as well as its morphologic mimics.

Case report

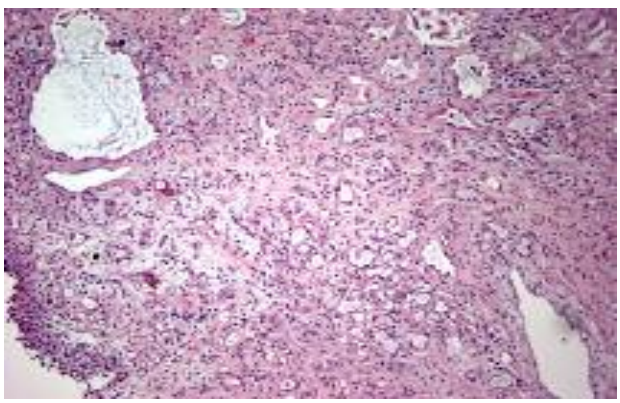
A 46-year-old female was seen for a consultation for intermittent painless gross hematuria for more than 1 month. She had a history of nephrectomy (11 years ago) for staghorn stone with non functioning left kidney, the pathological diagnosis concluded to renal tuberculosis. She was treated with antituberculosis drugs for 8 months.

Physical examination was normal. So were biological routine tests and urine analysis.

An ultrasound revealed a small lesion on the right side of the bladder. Cystouretroscopy disclosed diffuse, papillary, exophytic lesions over the trigone and all urinary bladder walls. A complete transurethral resection of the bladder tumors was performed.

The histological study revealed a tubular proliferation with rare edematous polyps. The tubules are small round structures lined by cuboidal epithelium (Fig. 1).

Figure 1 : Microscopic findings of nephrogenic adenoma show tubular structures lined with hobnail cells (H.& E. stain, × 100).



Cystic dilatation of the tubules is observed. Tubules and cysts contain focally eosinophilic secretions “colloid like” (Fig. 2).