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Case Report

Multiple Tumoral Calcinosis in a Hemodialysis Patient

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ABSTRACT

Pseudotumoral calcinosis is a rare condition characterized by periarticular calcium deposition. It preferentially affects large joints such as the hip, shoulder and elbow. It is a severe complication of chronic renal failure. We report a case of secondary pseudotumoral calcinosis in a chronic hemodialysis patient.

Keywords
Tumoral calcinosis; Chronicrenal failure; Hemodialysis.

INTRODUCTION

Tumoral calcinosis is a rare benign condition characterized by deposition of calcium salt in different peri-articular soft tissue regions, mainly in the juxta-articular areas.[1] It affects mostly African people, especially in the second decade, but it can affect any age.[2-4] The term tumoral calcinosis was first used by Inclan et al in 1943,[5] however, the condition itself was described before in 1899 by Duret.[6]

CASE REPORT

A 58-year-old patient, on chronic hemodialysis, presented with a six-month history of three slightly painful swellings gradually increasing in size located in the shoulder and both the hips. Clinical examination found a mass on the right shoulder (Figure 1) from which there was a flow of thick yellowish-white liquid. It also revealed the presence of three large lumps. The largest one on the right hip (measuring 20 cm in length) extended to the lower third of the thigh and was of firm consistency (Figure 2).

X-ray of the shoulder showed a periarticular calcified mass of multi-locular appearance capping the humoral head without lysing the bone. Magnetic resonance imaging (MRI) revealed the presence of several soft tissue masses on both shoulders and both hips, giving a bees nest-like appearance (Figure 3). Their matrix was osteoid and they were seen to infiltrate adjacent muscle whilst sparing bones and joints. The radiological findings were very suggestive of pseudotumoral calcinosis.

Laboratory tests showed a high serum calcium concentration, a normal phosphate concentration and elevated serum parathyroid hormone concentration. After the diagnosis of hypercalcemia with tumoral calcification secondary to chronic renal failure was made, the patient was placed on a low calcium diet and intensive hemodialysis sessions. However, the calcifications did not improve and the patient refused surgery.

Figure 1. *Mass of the Right Shoulder Showing Discharge of Thick Yellowish-White Liquid*

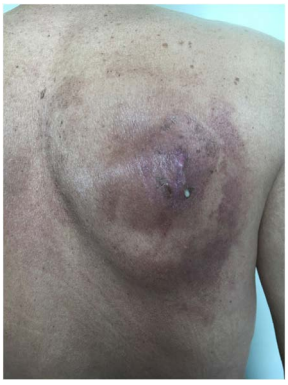
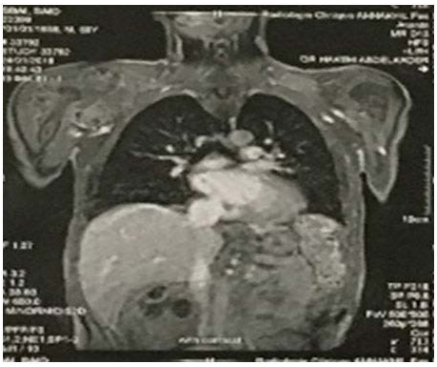


Figure 2. *Voluminous Mass, Measuring 20 cm on the Right Tigh and Extending above the Knee, of Firm Consistency*

Figure 3. *MRI Showing Soft Tissue Masses on both Shoulders, Giving a Beesnest-like Appearance*



DISCUSSION

There are three clinical forms of pseudotumoral calcinosis.

1. Tumoral calcinosis secondary to chronic renal failure due to a disorder in calcium and/or phosphate metabolism.
2. A familial form with a an autosomal recessive transmission, which may be hyper phosphataemic or normo-phosphataemic.
3. A form with a sporadic pattern which generally has normal calcium and phosphate levels.[7,8]

Although, lesions usually appear around large joints, cases involving cervical and lumbar spine, the supraclavicular area and toes have also been reported.[9] Large lesions can cause pain, functional impairment and nerve compression.[10] Local complications include skin ulceration with a chalky milk-like discharge and chronic infection with resulting secondary amyloidosis. Typically, there is no bone involvement, although periosteal reaction or erosion due to pressure may be found. Tumoral calcinosis has a typical appearance on radiographs: amorphous, cystic, and multilobulated calcification located in periarticular soft tissues. On MRI, tumoral calcinosis is seen as a well-circumscribed multicystic mass.[11] The differential diagnosis of tumoral calcinosis includes multiple causes of metabolic and dystrophic calcification including:

- Connective tissue diseases (systemic sclerosis, mixed connective tissue disease, dermatomyositis, systemic lupus erythematosus)
- Neoplastic diseases (synovial sarcoma, osteosarcoma, chondrosarcoma)
- Degenerative diseases (calcific tendonitis, calcific bursitis).[12]

Surgery is the gold standard treatment. Indications for surgery are pain, recurrent infection, ulceration and functional impairment.[13]

CONCLUSION

The diagnosis of tumoral calcinosis is based on clinical context, biology and radiology. Management of tumoral calcinosis is difficult. Surgical excision is the best treatment option, but the deposits can recur after surgery. Other options include increasing dialysis dosage phosphate deprivation and vitamin-D analogs but the response may be limited. Renal transplantational ways leads to resolution. However, more studies are necessary in order to clarify the most effective treatment modality for each type of tumoral calcinosis.[14]

CONSENT

The authors have received written informed consent from the patient.

CONFLICTS OF INTEREST

The authors declare that they have no conflicts of interest.

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Case Illustration

Genital Psoriasis: A Case Illustration

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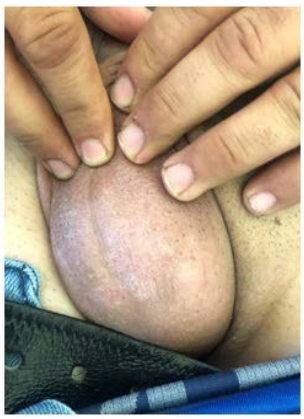
We present the case of a 35-year-old patient who has been suffering from a slightly pruritic erythematous lesion on the scrotum for 3-years (Figure 1). He was previously diagnosed with eczema and put on topical steroids with good improvement but later it recurred. On clinical examination, a finely squamous erythematous plaque was present on the scrotum without any other lesions elsewhere on the body. Dermoscopic examination showed regularly distributed red dots and a reddish background.

Figure 1. *On Clinical Examination, a Finely Squamous Erythematous Plaque*

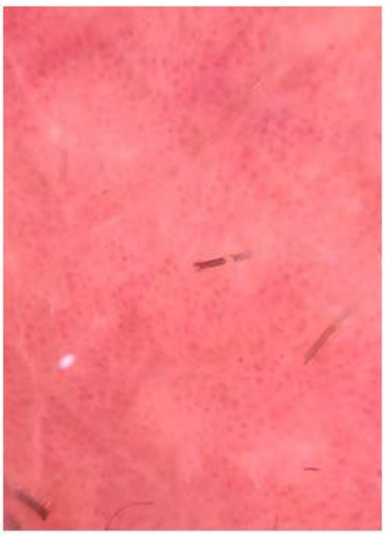


The diagnosis of psoriasis was confirmed and the patient was treated with topical steroids.

Psoriasis is a chronic, inflammatory epidermal skin disease with a high prevalence in the general population of approximately 2%. It is part of a more generalised plaque psoriasis, although the external genitalia may be the only area affected. However, the isolated presentation of psoriasis solely on genital skin seems to be rare and occurs in only 2-5% of the psoriatic patients. Prevalence is higher in men than in women and just few studies have been published on this topic.[1,2]

Lesions mainly affect the glans and the inner side of the fore skin (prepuce), and less frequently the sheath of the penis and the scrotum. Clinically, it manifest as erythematous plaques with white scales, rarely itching and it is the dermoscopic examination that has all its relevance to confirm the diagnosis. This shows regularly distributed dots on a reddish background (Figure 2).[3]

Figure 2. *The Dermoscopic Examination Showing Regularly Distributed Red Dots and Reddish Background*



CONSENT

The authors have received written informed consent from the patient.

CONFLICTS OF INTEREST

The authors declare that they have no conflicts of interest.

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Case Illustration

Necrosis of the Penis Due to an Electrocution: An Exceptional Case

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doi: 10.17140/UAOJ-4-123

INTRODUCTION

Electrocution accidents are defined by the passage of an electric current through the body. It usually takes place at home or in a professional setting. The analysis of the lesions at different affected systems (such as, cardiovascular, respiratory, cutaneous, neurological, and renal systems) which has affected in this case has allowed us to discover an unusual exit point.

OBSERVATION

A 36-year-old patient was presented with no notable pathological history who was victim of an electrocution accident by a high voltage electric current during the exercise of his profession as a house-painter. The initial examination in the intensive care unit found an unconscious patient, with a blood pressure at 100/50 mmHg, a tachycardia and a full bladder. The skin lesions were in the form of 2nd and 3rd-degree burns, occupying approximately 28% of the body surface and involving the internal sides of the thighs and the upper limbs. The urogenital examination found exposed scrotal tunics with necrosis of the penis and a retraction of the urethra (Figure 1). The treatment provided to the patient was based on resuscitation measures in order to maintain and support his vital functions. A suprapubic catheter was put to drain the urine. Afterward, he was admitted to the operating room for fasciotomy, treatment of burns and repair surgery of the genital organs. Unfortunately, he died from a cardiogenic shock.

Figure 1. *Photo of the Genitals Showing Exposed Scrotal Tunics (Red arrow) with Necrosis of the Penis (Green arrow) and a Retraction of the Urethra (Yellow arrow).*

DISCUSSION

The energy produced by the high voltage current is often so high that it causes severe burns. Most often, injuries sit in the point of contact or in the current's exit. A patient can have multiple entry and exit points. The most affected body parts are hands and head. As for the exit area, it is most often the heels.[1,2] Indeed, the genitals are an unusual exit point.[3,4] As in the case of traumatic or surgical amputation, penis reconstruction can be indicated in the case of penis necrosis secondary to an electrocution.[3] The reconstructive surgery aims to have an adequate length phallus with an apical meatus allowing satisfactory urinary and sexual functions.[5] Among the several techniques described, the penile lengthening by partial mobilization of the residual corpora cavernosa and spongiosa with covering by scrotal skin seems to allow the best results on the sensory and aesthetic level.[3,5,6]

CONCLUSION

High voltage electric current can cause serious injury, including death. Among these injuries, necrosis of the penis as found in this case is extremely rare. The treatment consists on a phalloplasty

with urethroplasty to insure urinary and sexual functions.

CONSENT

The authors have received written informed consent from the patient.

CONFLICTS OF INTEREST

The authors declare that they have no conflicts of interest.

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Case Report

Bilateral Pubo-Penile Ectopic Testis: A Case Report

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ABSTRACT

Pubo-penile ectopic testis is a rare congenital anomaly whose etiopathogenesis is still poorly understood. We report one case of bilateral pubo-penile ectopic testis in a two-year-old child. The diagnosis was made on physical examination alone. Orchidopexy in the dartos muscle was easy to perform.

Keywords
Pubo-Penile ectopic testis; Orchidopexy.

INTRODUCTION

Testicular ectopia (ectopic testis) is an aberrant migration of testis and so the testis is not found in the normal route of testicular descent into the scrotum. The testis therefore deviates from its usual path of descent.[1] The diagnosis is essentially clinical and the treatment is orchidopexy in Dartos.

CASE REPORT

A two-year-old boy was referred by his parents to the clinic for the management of bilateral absence of testis in the scrotum. Physical examination revealed a bilateral emptiness of the scrotum and a palpable mass corresponding to the ectopic testes in the pubo-penile region bilaterally (Figure 1). His general examination was normal. No additional diagnostic investigations were performed to confirm the diagnosis. At operation, the testes were approached by a supra-pubic skin-crease incision. During the surgical exploration, testes were found to be attached to the spermatic cord and otherwise normal (Figure 2). Bilateral orchidopexy with the testes transposed to bilateral pouches in the dartos muscle of the scrotum was performed without difficulty (Figure 2). Post-operative recovery was uneventful. The child was doing well on subsequent follow-up.

Figure 1. *Physical Examination and Surgical Exploration Revealed Emptiness of Scrotal Content and a Palpable Two Ovoid Swellings Localized to the Pubic Region*

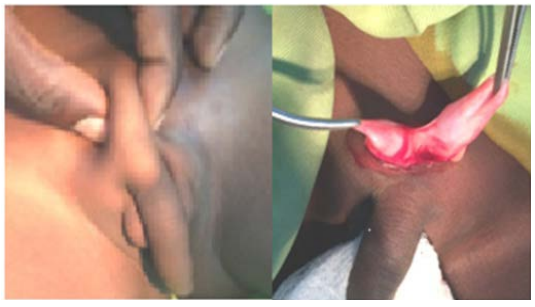
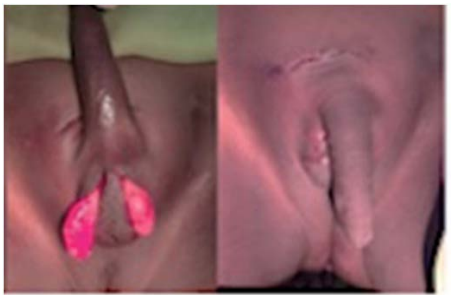


Figure 2. *Orchidopexy in Dartos*



DISCUSSION

An ectopic testis (testicular ectopia) is a rare congenital anomaly with an incidence of 1.5%.[2-6] Several clinical forms have been

reported based on anatomical position[2,7,8]:

- Position of the ectopic testis is at the superficial inguinal pouch
- Pubic type (position of the ectopic testis may be found at the root of the penis)
- Penile type (at penis)
- Perineal type (position of the ectopic testis may be found at the perineum)
- Femoral type (position of the ectopic testis is at the upper and at the medial part of the femoral triangle).

The etiopathogenesis of testicular ectopia is controversial.[1,8] The gubernaculum testis and testosterone are reported to be involved in the normal positioning of the testis.[3,4] According to Hutson et al,[5] gubernaculum testis weakness and an anomaly of the genito-femoral nerve could be the cause of aberrant migration and the occurrence of certain testicular ectopic positions. The genito-femoral nerve has a significant role as a tractor and guide of the gubernaculum.[9] For some authors. Kaufma [1,6] The Pubo-penile ectopic testis may be due to a mechanical obstruction at the level of the scrotum leading deviation of the testis towards a zone of less resistance. The case we report could possibly be due to this.

The diagnosis of pubic testicular ectopia is essentially based on physical examination and should be done at birth. For some authors, an ultrasound scan can be performed to confirm the diagnosis and aid localization. Doppler-associated ultrasound can demonstrate testicular vascularization.[4] In some cases, as recommended by Pugach et al,[10] pre-operative laparoscopies may be useful for diagnosis. Treatment is surgical[10] taking great care to avoid the risk of microtrauma and torsion so as to reduce the risk of ischemia and later fertility disorders. Some authors recommend performing orchidopexy as soon as possible before the age of one year.[10-12] However, delayed management is often reported.[1,6,7,10]

The long-term prognosis for pubo-penile ectopic testis is excellent because of the discretion of histological lesions. Hutcheson, on the other hand, in a comparative study, did not find a significant difference between the histological lesions observed in ectopic testis and cryptorchid testes.[9]

CONCLUSION

Pubo-penile ectopic testis is rare. The diagnosis is based on physical examination. Orchidopexy should be performed at the earliest possible opportunity.

CONSENT

The authors have received written informed consent from the patient.

CONFLICTS OF INTEREST

The authors declare that they have no conflicts of interest.

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Case Report

Collecting (Bellini) Duct Carcinoma: A Case Report of a Rare Tumor and Review of the Literature

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ABSTRACT

Renal cell carcinoma of the collecting ducts is one of the least frequent variants of renal carcinomas, with highly aggressive behavior, having the worst prognosis and the lowest specific cancer survival rate of all renal carcinomas, as 70% of patient deaths are secondary to the disease. We present a clinical case of a male patient with a diagnosis of paraneoplastic syndrome secondary to renal neoplasia. After a renal biopsy returned a diagnosis of sarcomatoid carcinoma, the patient elected surgical excision, and final pathology was consistent with renal carcinoma of the collecting ducts. As was common in the largest published series, this patient developed local and distant relapse in the early post-operative period, despite adjuvant systemic treatment. This variant of renal carcinoma has an ominous short-term prognosis, with high rates of distant disease present at the time of diagnosis. The unfavorable biological behavior manifests despite the use of multi-modality, adjuvant treatment.

Keywords
Bellini disease; Collecting ducts carcinoma; Renal carcinoma.

INTRODUCTION

Collecting ducts carcinoma or bellini duct carcinoma (CDC) is one of the most uncommon variants of renal carcinomas.[1] During last decade, no more than 20 cases have been reported and published in the United States (USA),[2,3] and a literature review identified 270 cases over the past 20-years.[4] Recently, Sui et al[5] published a report of 577 patients which represents the largest cohort of CDC in the literature to date CDC demonstrates highly aggressive behavior, being the variant of renal carcinoma with the worst prognosis and the lowest cancer-specific survival rate,[6] as 70% of deaths are secondary to the disease.

Early diagnosis is the main prognostic factor. However, most cases present with distant disease at the time of diagnosis.[7] Surgical treatment leads to the highest survival rates, especially small or confined (pT1) tumors. Different treatment protocols have been published, including chemotherapy, radiotherapy and multi-modality therapy, but without favorable responses in the majority of patients.[8]

The objective of this study is to report a case of CDC which presented with paraneoplastic syndrome, which was surgically treated and required systemic adjuvant treatment. In addition, a literature review of this disease also provided.

CASE REPORT

A 59-year-old male patient with past history of renal lithiasis and hyperuricemia presented with new onset asthenia and adynamia accompanied by night sweats, fever (<37.9 °C) and unintentional weight loss of 8-10 kg over 30-days. He denied abdominal pain, lower urinary tract symptoms, cough or other symptoms.

Physical examination was essentially unremarkable except for pale mucous membranes. Serum profile showed anemia, high leukocyte, neutrophil and platelet count, and increased acute phase reactants. Hematocrit 23.8%, Hemoglobin 7.4 gr/dL, leukocytes: 21.700 ml/mm^3, Platelets: 700,000 ml/mm^3, Erythrocyte sedimentation rate (ESR) 9 mm, C-Reactive Protein 129 mg/L, Glucose 132 mg/dL, Urea 32 mg/dL, Ionogram (sodium/potassium/chloride): 130 mEq/L / 4.2 mEq/L / 95 mEq/L, alkaline phosphatase level (ALP) 248 IU/L, Creatinine 1.12 mg/dL.

Urinary sediment, human immunodeficiency virus infection (HIV), blood and urine culture were unremarkable. Ultrasound and abdominal computerized tomography (CT) scan (Figures 1 and 2) revealed a 12×5 cm solid mass with heterogeneous enhancing soft tissue density, involving the left renal perihilar region and a accompanying left renal vein invasion with hilar compression level.

Figure 1. *Abdominal and Pelvic CT (Coronal Reconstruction). Tumor Lesion of 12 cm in Diameter at Cephalocaudal Level and 5 cm in the Axial Plane. Invasion of the Renal Vein (Direct Invasion and Proximal Stenosis in Topography of the Renal Hilum)*

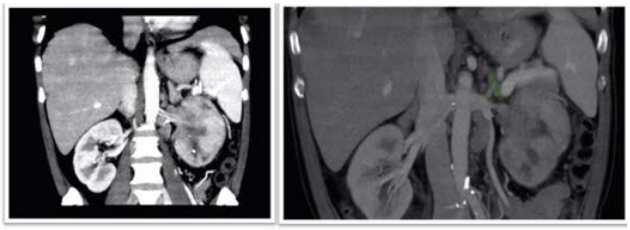


Figure 2. *CT Scan (Axial View): Intimate Contact with the Quadratus Lumborum, Muscle and to a Lesser Extent with the Upper 1/3 of the Illopsoas Muscle. Left Paraaortic and Perihiliar Lymph Nodes.*

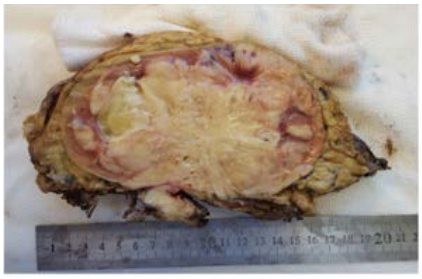
The mass was observed to extend posteriorly to contact the quadratus lumborum muscle and upper 1/3 of psoas, without a clear plane of separation. Multiple left perihilar and para-aortic lymph nodes were visualized (some greater than 2 cm). Renal morphometry score=12 p.

The clinical picture was interpreted as paraneoplastic syndrome secondary to renal neoplasia. Transfusions with red blood cells were indicated following invasive treatment. A coaxial-trucut (17 G coaxial needle and 18 G trucut sheath) biopsy was performed showing infiltration by undifferentiated fusocellar and pleomorphic neoplasia. Immunohistochemistry stained positive for renal cell carcinoma (RCC), cytokeratin (CK)7, CD10 binding, with biphasic phenotype consistent with sarcomatoid carcinoma.

The patient opted for surgical treatment which was performed *via* xiphoid-pubic midline incision. Radical nephrectomy with left para-aortic lymphadenectomy was performed with dissection of the retroperitoneum from the left iliac bifurcation to the left diaphragmatic pillar including the proximal and mid-left ureter. The post-operative course was uneventful, with hospital discharge on post-operative day (POD) number.[4]

On gross pathologic analysis, a grayish white tissue tumor measuring 12.5×9 cm was described. The tumor invaded the renal sinus and hilarfat, but did not extend through Gerota's fascia (Figure 3).

Figure 3. *Macroscopy: Renal Mass (12.5×9 cm in Diameter)*



Microscopy (Figure 4) revealedun differentiated sarcomatoid carcinoma (90%) with 50% necrosis, Fuhrman grade 4. Margins were negative. Lymphadenectomy showed eleven normal lymph nodes, thuspathological stage: pT3a N0

Figure 4. *Microscopy: Carcinoma with a Sarcomatoid Phenotype (Spindle Cell)*

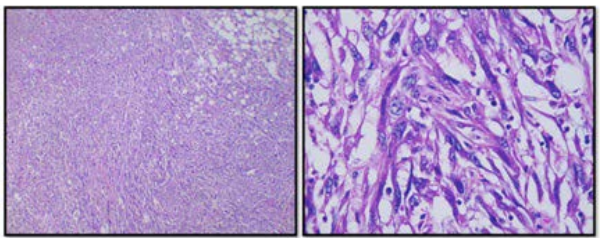


Figure 5. *Inmunohistochemistry (IHC): CD10 - positive and CD7 - positive*

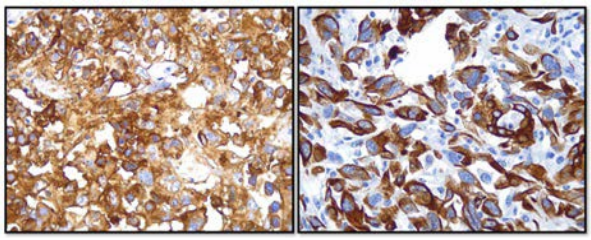
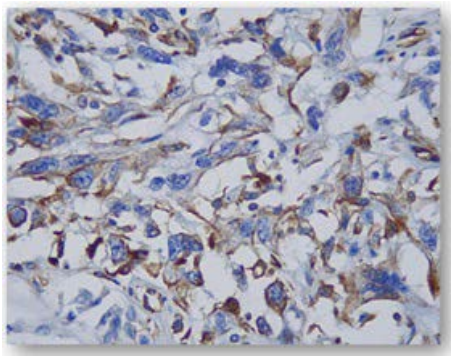


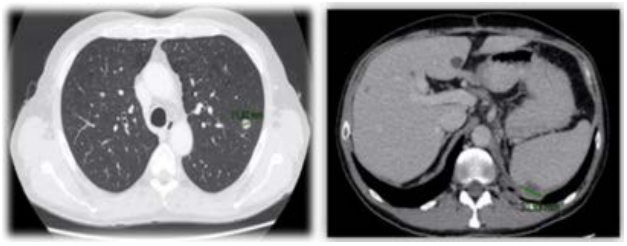
Figure 6. *IHC:Vimentin Positive*



Immunohistochemistry shown in Figures 5 and 6, also stained positive for Vimentin CD 10, CK7 and CK19, and negative for p53. CDC with sarcomatoid features was the final histologic diagnosis.

During the fourth month of follow-up, a chest and abdomen CT scan showed a 12 mm nodular lesion in left superior pulmonary lobe and a 31 mm lesion in upper pole of the spleen just below the diaphragm, consistent with metastases (Figure 7).

Figure 7. *Chest/Abdomen/Pelvis CT Scan (Following Initial Surgical Treatment): Local Recurrence and Lung Metastasis*



A biopsy of the sub-diaphragmatic lesion was performed using a coaxial-trucut system showed recurrent fusocellular and epitheloid neoplasia, with extensive necrosis.

The patient is now receiving a second cycle of adjuvant chemotherapy (Cisplatin-Gemcitabine) without significant adverse events.

DISCUSSION

In 1976, Mancilla-Jiménez et al published the first CDC case, which reported the hyperplastic and atypical changes adjacent to epithelium of collecting ducts in 3 of 34 cases of renal carcinoma.[9] The embryological origin differs from other renal parenchymal tumors. The renal collecting system (ureters, pelvis, calices and collecting ducts) are derived from the ureteric bud, which originates from the Wolffian duct, while the renal parenchyma derives from the metanephric blastema. This explains the different clinical, radiological, macroscopic, microscopic, immunohistochemical and cytogenetic characteristics of CDC *vs* RCC.[10]

According to the conference of International Society of Urological Pathology (ISUP) on renal neoplasia in Vancouver in 2013, CDC must include at least some medullary lesions, have a predominant tubule formation, and have an inflamed desmoplastic stroma. They must have high-grade cytological features, infiltrative growth patterns, and lastly, no features compatible with other subtypes of renal carcinoma or urothelial carcinoma.[11] Immunohistochemistry, it is usually positive for high molecular weight cytokeratins (CK19, CK7, CK8/CK18), Fez1, mucin, lysozyme and lectins.[12]

Tokuda et al published the largest series and reported an incidence 0.4 to 1.8% of all renal tumors, with 70% prevalence in male and young adults (mean 58.2-years).[4]

More than a half of patients (65.4%) are symptomatic at the time of diagnosis. The most common presenting symptoms are hematuria and lumbar or abdominal pain, but patients with systemic symptoms such as fever of unknown origin, weight loss, or elevated acute phase reactants are not uncommon. Patients with systemic (paraneoplastic) symptoms often have advanced or distant disease at diagnosis when compared with other types of renal carcinomas.[13] More than 75% of CDC present with pT2-pT3 disease with 95% of nuclear grade.[4]

The most common sites of metastasis are regional lymph nodes, lung and bone. In the Tokuda et al series, 44.2% of patients presented lymphatic metastasis and 32.1% distant metastases.

During the pre-operative evaluation, CT scans are not specific so there is difficulty differentiating from other subtypes of renal carcinomas. In some cases, reporting the use of angiography, CDC is typically hypovascular, while 90% of clear cell renal carcinomas are hypervascular.[14]

Prior reports observe cancer specific survival, of 69% at one year post diagnosis, falling to 45% and 34% in third and fifth-year respectively.[4] Ciszwesky et al[4] postulated that mean time of local recurrence and distant metastasis after nephrectomy was 4.9 and 8.1-months respectively.[8]

Attempts to control the disease with immunotherapy or chemotherapy have met with limited success. Chemotherapy for urothelial carcinomas is used because of mesonephric origin of CDC. The most common regimen employed has been methotrexate, vinblastine, doxorubicin and cisplatin.[4] However, the largest prospective series of 23 patients treated with gemcitabine,[15] demonstrate an overall response rate of 26% in metastatic CDC. Oudard et al[15] propose gemcitabine as first line treatment in stage IV CDC. In this phase II, multi-center trial, a combination of gemcitabine and cisplatin/carboplatin was utilized in 23 patients with metastatic CDC.15 Overall survival at 1-year was 48%, decreasing to 17% at 18-months. Tokuda, et al studied 34 patients treated with immunotherapy (interferon alpha and gamma and interleukin-2 regimen) and found no response.[4]

A retrospective review of 64 cases of metastatic, non–clear cell renal cell carcinoma reported on 26 cases of CDC. Of these patients, one had a 5-month partial response to gemcitabine plus cisplatin.[16]

Currently, Siu et al[5] reported 577 CDC patients with overall survival for the metastatic CDC cohort of 6.4-months. On sub-analysis, the utilization of surgery with chemo/radiation was associated with decreased risk of death (HR=0.51, 95% CI: 0.32-0.79) compared to surgery, alone, and also compared with chemo/radiation alone (HR=0.57, 95% CI: 0.37-0.89).

CONCLUSION

The collecting duct carcinoma of Bellini is an uncommon malignant

renal neoplasm variant which has a poor prognosis, due to high rates of distant disease at the time of presentation. In this case report, the typical biological behavior of local recurrence and metastasis within a few months after surgery was observed. Due to the rare nature and unfavorable outcomes in CDC, a cooperative group trial to assess possible neoadjuvant and adjuvant chemotherapy prior to and after surgical resection, should be considered.

DECLARATION

This case report is ethically approval by Institutional Ethical Committee.

CONSENT

The authors have received written informed consent from the patient.

CONFLICTS OF INTEREST

The authors declare that they have no conflicts of interest.

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