

The Tunica vaginalis testis metastasis as the first clinical manifestation of pancreatic adenocarcinoma: a case report and and literature review

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Case report

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Abstract

Background: Metastases from pancreas or ampullary malignancies are common, but spread to the testicle and paratesticular tissue is exceedingly rare. To our knowledge, few than 30 cases have been previously reported in the literature. More rarely, it is to be found with tunica vaginalis testis metastasis without involvement of testicle and epididymis.

Case presentation: A 65-year-male with complaints of left painless scrotal swellings for over 1 week was referred to the Department of Urology . Scrotal ultrasound demonstrated a left sided hydrocele with paratesticular masses. The chest computed tomography showed lung metastases and left enlarged supraclavicular lymph node. The preoperative diagnosis was left testicular tumor with lung metastasis. Then, a left radical orchidectomy was performed with high ligation of the spermatic cord and postoperative histopathology showed that it was suggestive of metastatic tumors. Abdominal computed tomography revealed the presence of a tumor in the tail of the pancreas. Tumor markers cancer antigen CA 19-9 were elevated almost 6-fold levels. The outcome of this patient was unsatisfactory and died 3 months later.

Conclusions: This case highlighted that metastatic carcinoma from the pancreas must be considered in the differential diagnosis of scrotal enlargement. Older patient age is suggestive of a secondary testicular tumor. In addition, careful clinical and radiological examination has become the imaging modality.

Background

Metastatic testicular or paratesticular tumors that are formed from another solid tumor origin are relatively uncommon, accounting for only 0.02–3.6% of all testicular neoplastic lesions [1]. The most common primary sites are the prostate, kidney, gastrointestinal tract, lung and breast [2]. In a review of the literature, we found fewer than 30 cases of testicular or paratesticular metastases arising from a pancreatic neoplasm. Here we present a case of tunica vaginalis testis metastasis from pancreatic adenocarcinoma in 65-year-old man and provide a comprehensive literature review with the objective of providing useful information on this malignancy. This case has a rare presentation: it shows that pancreatic adenocarcinoma in an adolescent patient can present with paratesticular metastasis as well.

Case Presentation

A 65-year-male was referred to the Department of Urology with left painless scrotal swellings, which was gradually increasing in size over 1 week. Physical examination revealed a 2 × 3 cm hard, globular mass with transillumination in the lower part of the left scrotum. His right testis was of normal size with no associated scrotal swelling. There was no palpable inguinal lymphadenopathy or abdominal masses. Scrotal ultrasound demonstrated a left sided hydrocele with paratesticular masses. He presented with dyslipidemia, hypertension and type 2 diabetes and had no history of scrotal trauma, no medical history of genitourinary tract anomalies and did not report any asbestos exposure or any surgical procedures. He denied anorexia, weight loss, fever, dysuria, history of STI, and recent sick contacts.

Pre-operative blood tests, including the β -subunit of human chorionic gonadotropin (β -hCG) and α -fetoprotein (AFP), and lactate dehydrogenase were all normal. The chest computed tomography showed lung metastases

and left enlarged supraclavicular lymph node (Fig. 1).

Clinically, the preoperative diagnosis was left testicular tumor with lung metastasis. A left radical orchidectomy was performed with high ligation of the spermatic cord. During the operation, multiple nodules with variable size were noted inner the surface of tunica vaginalis, while the left testis was not involved(Fig. 2). Final pathologic examination showed infiltration of malignant cells in the tunica vaginalis, which was suggestive of metastatic tumors (Fig. 3).

Taken together, such findings allowed us to rule out a primary tunica vaginalis cancer as well as metastasis from extratesticular carcinoma. Digital rectal examination, prostate specific antigen, showed normal findings. However, tumor markers cancer antigen CA 19 – 9 were significantly elevated. (203.1 U/mL; reference, 0–34.0 U/mL). Meanwhile, computed tomography of his chest, abdomen, pelvis, and brain was done as a part of metastatic workup and demonstrated the presence of a tumor of the tail of the pancreas and metastatic foci of the disease in the liver. Retroperitoneal lymphadenopathy, nodular infiltrates in the omentum and ascites were also found. There was no lesion in his brain suggestive of metastasis(Fig. 4). Based on these findings, the patient was diagnosed as having a metastatic tunica vaginalis carcinoma originating from pancreas with peritoneal carcinomatosis, retroperitoneal lymphadenopathy and lung metastasis. In view of distant metastasis, we referred to oncologist for further chemotherapy. But he denied any medical intervention. The entire course of illness from appearance of first symptom to death was only 3 months.

Discussion

Metastatic malignancies to the testicle and paratesticular tissue are extremely rare. The most frequent primary cancers are prostate, lung, kidney, gastrointestinal tumors and breast cancers [2]. In particular, very limited cases reported metastasis from pancreatic cancer to the testicle and paratesticular tissue. Kiefer ED first described metastatic epididymal spread from primary pancreatic carcinoma in 1927 as an incidental autopsy finding [3]. To date, less than 30 cases have been reported, including one from China [4], two from South Korea [5–6], ten from Japan [7], six from the USA [8–12] and the other five from European countries [13–17]. The scrotal or inguinal metastasis propensity occurs world-wide, although there is a remarkably high incidence among Japanese men, who account for one half of published cases. For this literature review, we searched relevant case reports that were available in full-text. Some cases that did not contain detailed information on treatment and prognosis were excluded. Consequently, a total of 15 cases documented in 14 published papers were included in our review (Table 1).

Table 1 Reports of metastatic paratesticular or testicular tumors of pancreatic or duodenum cancer

Case No.	Age (y)	Symptoms	Duration	Site	size (cm)	Metastatic Organ	Treatment	Primary location and histopathology	Prognosis after treatment
1[4]	65	Painless scrotal swelling	9 months	left		Left testis Right lung	Radical orchiectomy+ Pancreatic mass resection+ right lung tumor biopsy	Pancreatic body adenocarcinoma	Alive,9 month
2[5]	67	Painless scrotal swelling	3 months	left	7 × 5	Left paratestis Peritoneum,bone	Radical orchiectomy+gemcitabine chemotherapy	Pancreatic tail Mucinous cystadenocarcinoma	Died,3 month
5[6]	69	Painful scrotal swelling	NA	left	NA	Tunica virginals testis Liver, peritoneum Omentum	Hydrocelectomy+ gemcitabine chemotherapy	Pancreatic tail Adenocarcinoma	NA
11[7]	58	Painful scrotal swelling,	NA	left	3-4	Epididymis,spermatic cord,Stomach,Left kidney ,spleen	Radical orchiectomy+Pancreatic tumor biopsy	Pancreatic tail Adenocarcinoma	Died,3 months
6[8]	42	Jaundice,dark urine,pale stool, Painful scrotal swelling, Weight loss	3 weeks	left	NA	Omentum, tunica vaginalis testis, porta hepatis.	Exploratory laparotomy, left scrotal mass biopsy.	Pancreatic tail Adenocarcinoma	NA.
8[9]	53	Painful scrotal swelling	NA	right	4	Epididymis Liver	Radical orchiectomy+ Pancreatic tumor biopsy	Pancreatic head ,body and uncinete process Adenocarcinoma	Died,16 months
9[9]	36	Painless scrotal swelling,	18 months	right	NA	right testis,Epididymis Spermatic cord	Radical orchiectomy	Ampullary Adenocarcinoma	Died,2 months
10[10]	58	Painful scrotal swelling,	1 month	left	7.0× 4.5×3.5	Left testis, Liver,	Radical orchiectomy+ Pancreatic tumor biopsy	Pancreatic tail Adenocarcinoma	Alive,6 month
12[11]	70	Painless scrotal swelling,	21 months	right	NA	Tunica vaginalis testis	Hydrocelectomy Pancreaticoduodenectomy capecitabine chemoradiation	Ampullary Adenocarcinoma	Alive,1 month
15[12]	41	painful scrotal swelling	4 months	right	1.7,0.8	the spermatic cord epididymis	Radical orchidectomy+ chemotherapy	Pancreatic head and body Adenocarcinoma	Died,12 months
3[13]	36	Painful scrotal swelling	NA	right	NA	right testis, liver	Radical orchiectomy+ chemotherapy	Pancreatic tail.Adenocarcinoma	Died,3 month
4[14]	73	Painless scrotal swelling, Weight loss	NA	left	4 × 8	Left paratestis Liver, Lung, Retroperitoneum, Left suprarenal gland	Radical orchiectomy	Pancreatic Adenocarcinoma	Died,2 month
7[15]	70	Painless scrotal swelling,	NA	right	2	Epididymis Spermatic cord,liver	Pancreatic tumor biopsy+ orchifunicolectomy+ gemcitabine and abraxane chemotherapy	Ductal Pancreatic Adenocarcinoma	NA
13[16]	77	Painless scrotal swelling, Weight loss, Abdominal pain	1 month	right	3.0× 2.0	Right testis	Radical orchiectomy+ Pancreatic tumor biopsy	Mucinous exocrine pancreatic Adenocarcinoma	NA

14[17]	67	Mass in groin, recurrent vomiting	3 months	right		the spermatic cord duodenum	Radical orchidectomy Pancreaticoduodenectomy, splenectomy, chemotherapy	Pancreatic body and tail Adenocarcinoma	Alive, 4 weeks
Our case	65	Painless scrotal swelling,	1 week	left	2x3	Tunica vaginalis testis Liver, Omentum Retroperitoneum	Radical orchidectomy	Pancreatic tail Adenocarcinoma	Died, 3 months

Abbreviations □ NA, not available

Primary testicular tumors are usually diagnosed between the second and fourth decades, while secondary testicular tumors peak in the fifth and sixth decades. The mean age of incidence was 59 years (range: 36 to 77 years), and the peak incidence was in the 5th through 7th decades, with the similarity of the review reported by Tanaka H [7]. As the presentation of pancreatic cancer is often insidious, with nonspecific symptoms such as nausea and anorexia, which delay diagnosis until other more ominous symptoms such as weight loss, abdominal pain, or gastrointestinal symptoms develop. Our review showed that most testicular metastasis cases present as a palpable, painless or painful, slowly enlarging mass in the scrotum, which were easily neglected or misdiagnosed as primary testis lesion. There were only three cases who present with weight loss or other digestive system discomfort [8, 14, 16]. One patient was referred because of acutely developed severe pain of right testis [13], who was likely diagnosed as orchitis. Kim YW and colleagues reported a metastatic testicular tumor from pancreas, who presented only with hydrocele as initial symptom [6]. In the present case, the scrotal mass was the first clinical manifestation of the underlying malignancy. The average duration of onset was ranged from one week to 21 months. Hirano D and colleagues found that the bilateral testis were equally involved in metastatic tumors of the spermatic cord originating from 8 stomach cases, 8 colon cases, 2 liver cases, and 2 kidney cases in his review [18]. Tanaka H and colleagues revealed right testis was easily involved in metastatic tumors of the epididymis and the spermatic cord from pancreatic carcinoma, with the ratio 9 to 1 [7]. However, in our literature review, we found that the same occurrence in both sides in cases of scrotal or inguinal metastasis from pancreatic carcinoma. The average metastatic tumor size of in the identified 15 cases was 3.6 cm in a diameter (range: 1.6 to 6.5 cm). The tumor size ranged from 2.0 cm to 8.0 cm. Large tumor was reported in two cases [5, 10]. In addition, we discovered a significant feature that 8 cases of carcinomas originating from pancreatic tail were susceptible to metastasize to testis or paratestis compared with tumors from pancreatic head or ampulla.

The mechanisms of metastasis to the scrotal and inguinal tissues from primary malignant neoplasms have not been precisely elucidated. But it has been widely recognized that main routes include arterial embolization, transperitoneal seeding through tunica vaginalis. In our review, the metastatic tumors extending to the testis were found in five (31%) of the identified 16 cases [4, 9, 10, 13, 16], of which one cases invaded the epididymis [9]. Meanwhile, the tumors extending to the paratestis were reported in 11 cases [5, 6, 7, 8, 11, 12, 14, 15, 17], of which 4 cases involved the tunica vaginalis rather than testis [6, 8, 11], six cases invaded the spermatic cord and /or epididymis [5, 7, 12, 14, 15, 17]. Our case is unique since there were almost no symptoms of the primary tumor, only with paratesticular nodules, which proved to be metastasis and the first sign of pancreatic carcinoma. As there was obvious evidence of retroperitoneal involvement showed in CT scan, the suspected route of tumor spread in this case is either lymphatic or direct transperitoneal seeding from peritoneal carcinomatosis. Due to only one-week duration of onset, the right testicle was clear.

Clinically, no specific features that differentiate primary from secondary testicular or paratesticular tumors are available, as both may present with painful or painless mass, or an indurated testis. Serum tumor markers such as AFP and β -hCG are not helpful in distinguishing primary from secondary testicular tumors. However, one third of patients with pancreatic exocrine adenocarcinoma were found elevated β -hCG levels. Taylor H described a case of pancreatic adenocarcinoma presenting as a testicular tumor featured raised 10-fold hCG levels due to extragonadal secretion [16]. Immunohistochemistry is currently considered as the most sensitive and specific way of determining the origin of the tumor. CA19-9, CDX-2, cytokeratin are reliable markers. Another new markers, such as homeobox protein NANOG, SOX2, and Oct-3/4 have been applied in diagnosis [19–20]. In our case, pancreatic origin was suspected because of extremely elevated tumor markers CA 19 – 9 and positive CT results.

Most cases as well as our case underwent radical orchiectomy, while hydrocelectomy with preservation of the testis was found in two cases [6, 11]. One patient with omentum involvement was managed by scrotal mass biopsy [8]. About half of the cases received chemotherapy including gemcitabine or capecitabine. But the outcome was unsatisfactory with high mortality. In our literature review, two thirds of the patients died in their recorded follow-up period. The shortest survival duration recorded was only 2 months. Compared to other pancreatic origin, ampullary tumors have a higher rate of resectability and a more favorable prognosis. Our review shows that one metastatic ampullary adenocarcinoma patient who underwent pancreaticoduodenectomy had better outcome compared to another case who did not receive primary tumor resection.

Conclusions

Pancreatic cancer carries an unfavorable prognosis and given insidious symptoms has often metastasized at the time of presentation. We report a case of tunica vaginalis testis metastasis from pancreatic adenocarcinoma in 65-year-old man presented with painless swelling as the initial and solitary symptom. Neither situation has been described before in the literature. This case highlighted that metastatic carcinoma from the pancreas must be considered in the differential diagnosis of scrotal enlargement. Older patient age is suggestive of a secondary testicular tumor. In addition, careful clinical and radiological examination has become the imaging modality.

Declarations

Authors' contributions

YRZ, BSG and WA perform the surgery. YWF wrote the initial draft. KMG and DGM reviewed the literature. All authors have read and approved the final manuscript.

Availability of data and materials

The datasets generated and analyzed during the present study are available from the corresponding author on reasonable request.

Ethics approval and consent to participate

The study was approved by the Medical Ethics Committee of the First Hospital of Jilin University.

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Competing interests

The authors declare that there are no competing interests associated with the manuscript.

Consent for publication

All data published here are under the consent for publication. Written informed consent was obtained from all individual participants included in the study.

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Figures

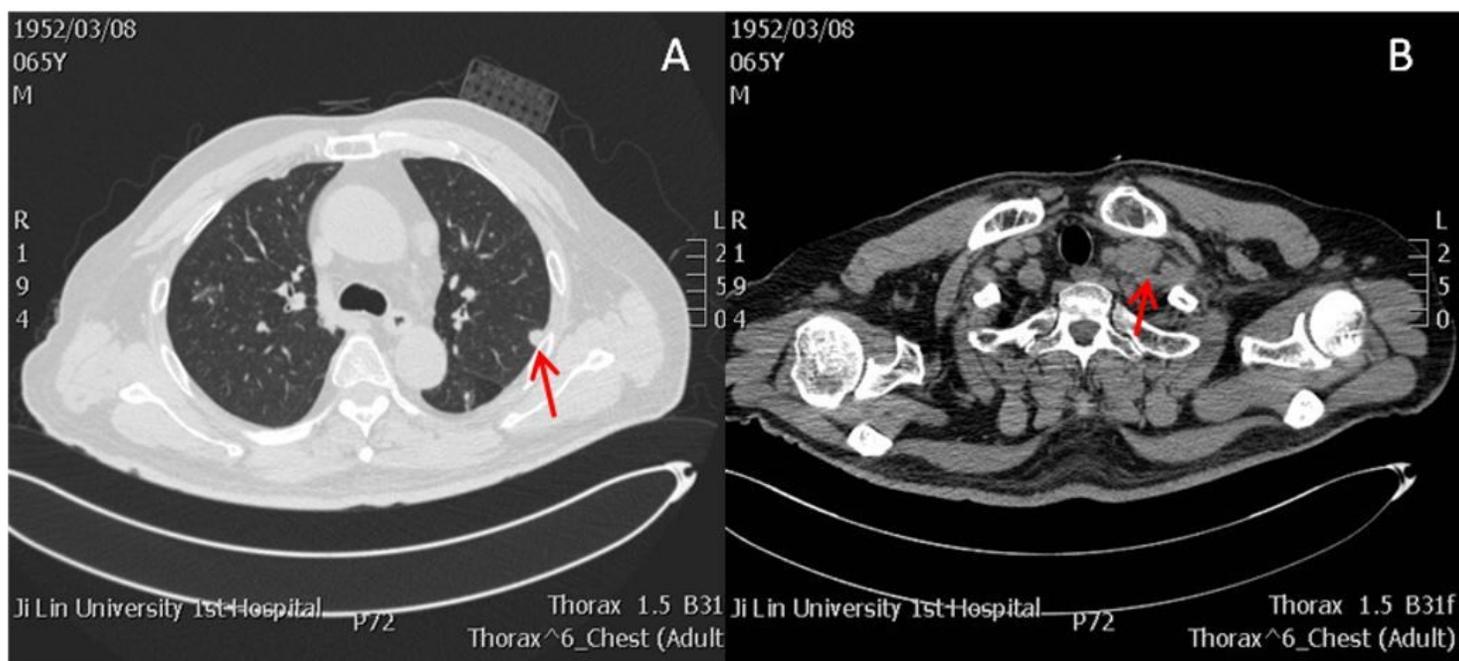


Figure 1

The chest radiologic findings. (A) A round mass in left lung lobe was observed.(B)Left enlarged supraclavicular lymph node was also noted. (Red arrow indicating relevant lesions)

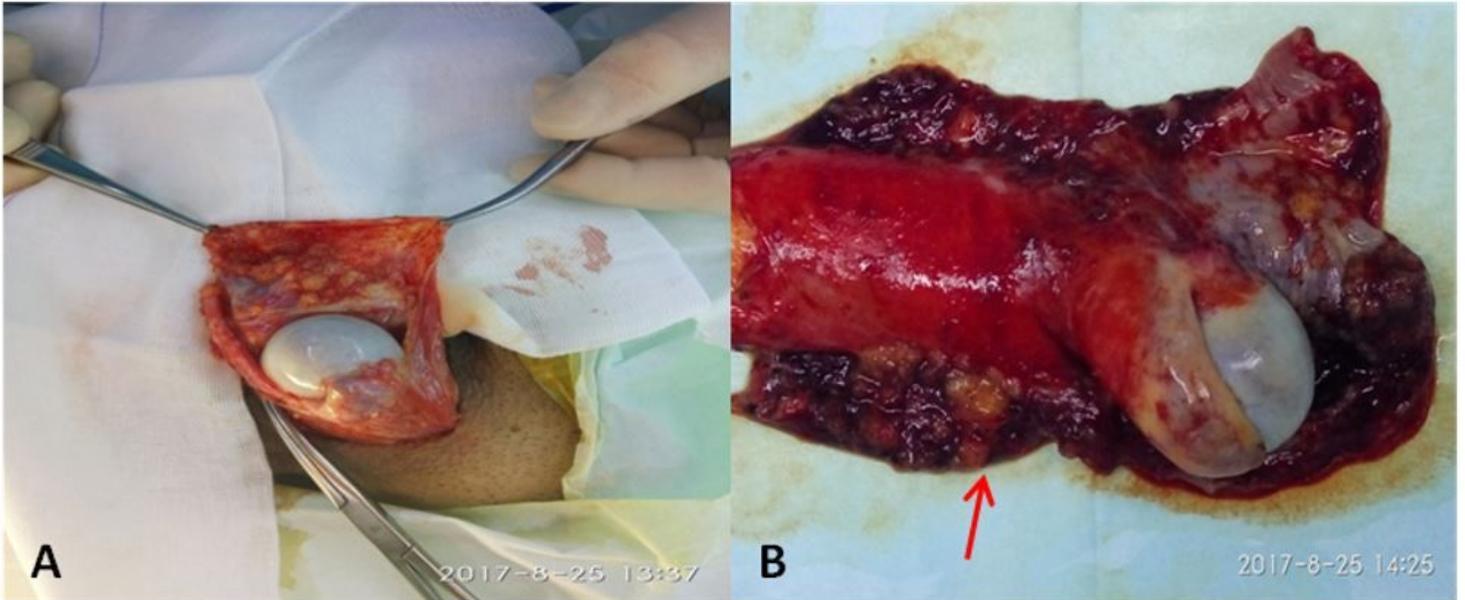


Figure 2

Gross appearance of metastatic tunica vaginalis tumor. (A) Left orchiectomy via inguinal canal approach was performed.(B)Macroscopic appearance of the surgical specimen showing multiple surface polypoid nodules in tunica vaginalis (Red arrow).

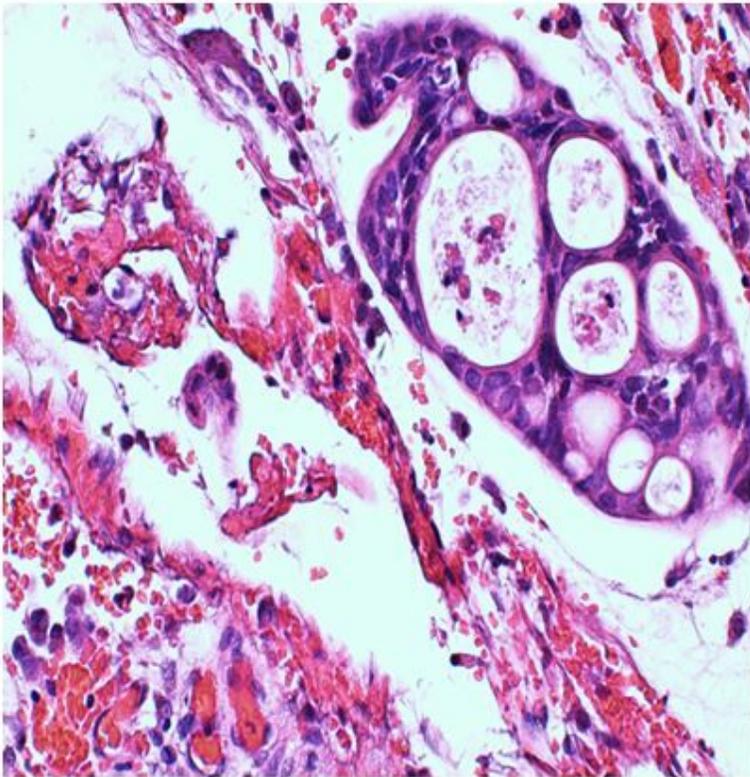


Figure 3

HE staining of metastatic tunica vaginalis tumor. Infiltration of malignant cells into tunica vaginalis tissue was observed. (H&E ×100).

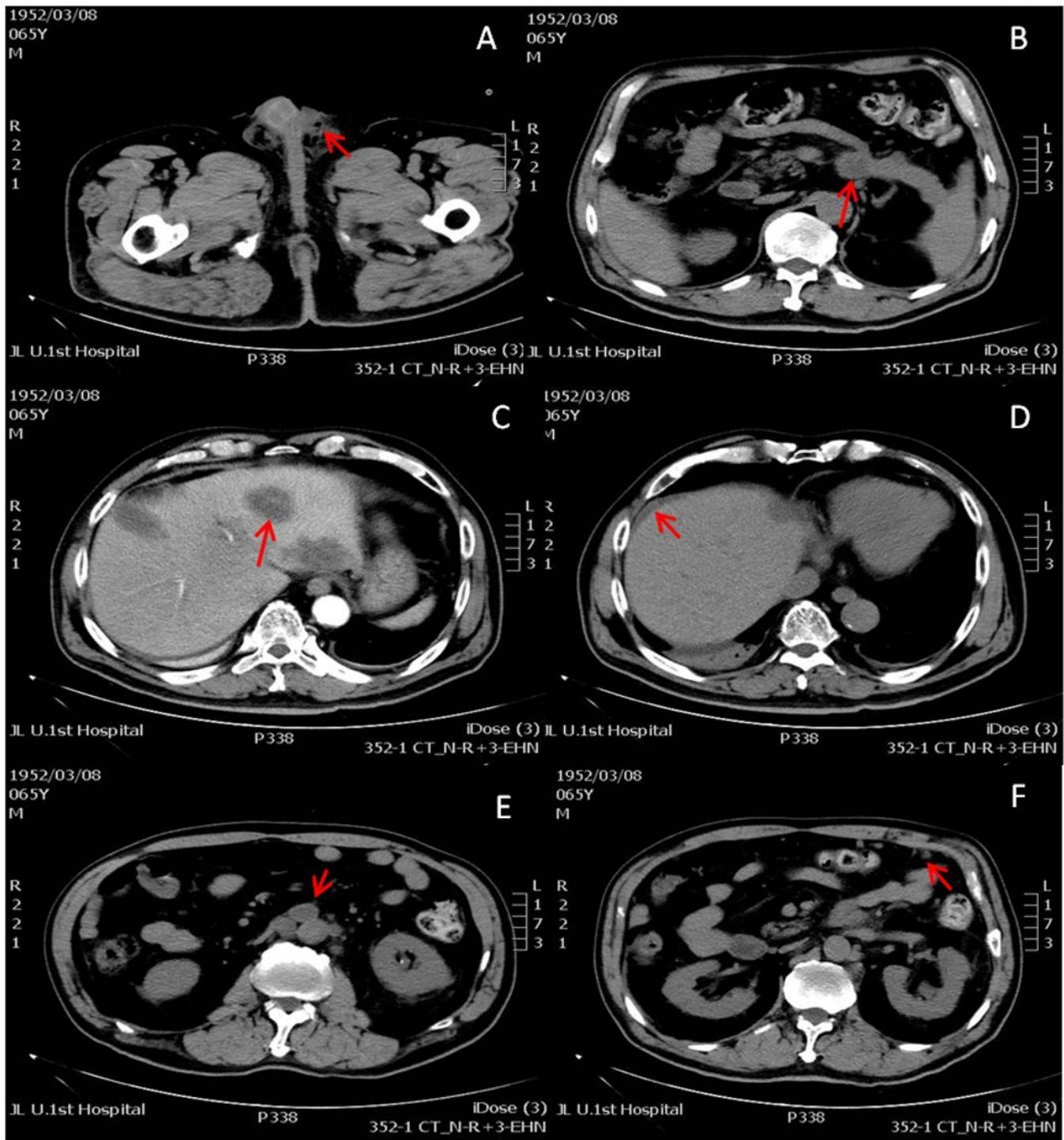


Figure 4

Postoperative Abdominal Radiologic findings (A) A left radical orchidectomy has been performed with high ligation of the spermatic cord. (Red arrow indicating air density spot). (B) primary tumor can be seen in the tail of pancreas.(C) multiple hepatic metastasis.(D) abdominal cavity fluid. (E) retroperitoneal lymphadenopathy. (F) omentum metastasis. (Red arrow indicating relevant lesions)