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Eosinophilic funiculitis initially diagnosed as irreducible inguinal hernia: A case report



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ABSTRACT

BACKGROUND: Most groin masses are first suspected to be groin hernias. More than 80% of bulging groin lesions are reportedly diagnosed as hernias by ultrasonography. Establishment of the correct diagnosis of hernia among all differential diagnoses is not easy. We herein describe a very rare case of groin eosinophilic funiculitis that presented as an irreducible groin hernia.

CASE PRESENTATION: A 59-year-old man presented to our hospital with suspicion of a right groin hernia. He had a 1-week history of a painful right groin tumor. The tumor was about 4 cm without skin redness or warmth, irreducible even in the supine position, and associated with mild tenderness. Enhanced computed tomography showed that the mass seemed to be connected to the intra-abdominal structures. With time, the patient's pain did not increase, the inflammatory response did not worsen, and no ischemic signs were observed by enhanced computed tomography. Therefore, we diagnosed the tumor as an irreducible but not incarcerated hernia and performed elective surgery. Intraoperative examination revealed no hernia sac, and a 4 × 3-cm tumor was observed around the spermatic cord. A malignant tumor was not completely ruled out. High orchiectomy was performed after consultation with the urologists. Pathological examination of the tumor showed no malignant features, and the final diagnosis was eosinophilic funiculitis with massive inflammatory changes and eosinophil invasion.

CONCLUSION: Eosinophilic funiculitis is very rare; only three cases have been reported to date. We should always consider unusual causes of groin masses during a surgical approach to hernia-like lesions.

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1. Introduction

One of the most common diagnoses for groin bulging is a groin hernia, which is reportedly present in 84% of such cases as shown by

Abbreviations: CT, computed tomography; IgE, immunoglobulin E; ANCA, anti-neutrophilic cytoplasmic antibody.

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ultrasonography [1]. Other differential diagnoses, although uncommon, are inflammatory lesions and malignant neoplasms [2–4].

Well-known inflammatory diseases associated with local eosinophilic infiltration are eosinophilic pneumonia, eosinophilic digestive disease, and eosinophilic chronic rhinosinusitis [5–7].

We herein report a very rare case of eosinophilic funiculitis that was preoperatively diagnosed as an irreducible groin hernia, illustrating how surgeons or clinicians may mistake when the cause of an inguinal mass is one of these less common etiologies. This report is based on Surgical Case Report (SCARE) Guidelines [8].

2. Presentation of case

A 59-years-old man was referred to our hospital for evaluation of right groin bulging. His height and weight were 159.0 cm and 66.2 kg respectively. His medical history included hypertension, hyperlipidemia, hyperuricemia, and dermal pruritus, and his child had atopic dermatitis. He felt upper abdominal pain and nausea and



Fig. 1. Abdominal enhanced CT. The right groin mass (arrow) was connected from the abdominal cavity to scrotum which suggested right groin hernia (A; sagittal section, B; coronal section). The fatty tissue around the stomach was enhanced and lymphadenopathy (arrow head) around the stomach, para aorta and mesentery was shown (C).

had noticed the right groin mass with mild pain after heavy drinking 1 week previously. He was diagnosed with acute pancreatitis and medicated. The groin mass did not change in size, so he was referred to our hospital with suspicion of an irreducible inguinal hernia. The patient was afebrile. His abdomen was not distended and was soft with no tenderness. The right groin mass was about 4 cm without redness of the skin. It was elastic hard and mildly tender, had no localized warmth, and was manually irreducible.

In the laboratory data, lactate dehydrogenase (317 IU/L; normal range, 119–229 IU/L), C-reactive protein (1.0 mg/dL; normal range, 0.0–0.5 mg/dL), creatine kinase (372 IU/L; normal range, 62–287 IU/L), and the erythrocyte sedimentation rate (45 mm/h; normal range, 2–10 mm/h) were slightly elevated. All other blood parameters, including the white blood cell, neutrophil and eosinophil counts were within normal limits.

Abdominal enhanced CT showed that the right groin mass seemed to be connected from the abdominal cavity to the scrotum, suggesting a groin hernia (Fig. 1). The hernial contents were considered to be fatty tissue with no sign of ischemic change or strangulation, but CT showed inflammatory change in the inguinal canal, so we consult to the urologists for suspicious of testicular torsion, then it was negative by ultrasonography of testis. Lymphadenopathy around the stomach, para-aortic tissue and mesentery was also observed. The patient had undergone examination using a gastrointestinal camera with his family doctor, and no gastric cancer was observed at that time. Based on these findings, we diagnosed the patient with an irreducible but not incarcerated right groin hernia and performed elective surgery.

The operation was begun with anterior approach. After dissecting the inguinal canal, the hernia sac could not be found. An approximately 4-cm elastic hard tumor surrounded and tightly adhered to the spermatic cord (Fig. 2). After consultation with the urologists, a malignancy was strongly suspected. Preoperatively, we had provided an insufficient explanation to the patient and his family that the groin tumor might be a malignancy, and we did not obtain informed consent from them for an additional operative procedure involving resection of the spermatic cord and testis. Therefore, during the operation, we fully explained to his family that there was a strong possibility of a malignancy and the need for tumor resection including the spermatic cord, testicular vessels, and testis. After obtaining informed consent from the patient's family, we performed high orchiectomy.

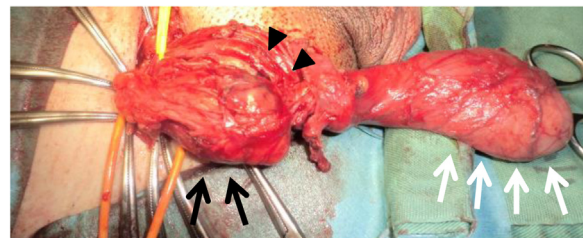


Fig. 2. Intraoperative findings. There was 4 cm elastic hard tumor (arrow) surrounding the spermatic cord (arrow head) and it adhered tightly, therefore the high orchiectomy was performed (testis; white arrow).

In macroscopic findings, a 4 × 3-cm tumor was located at the proximal of the spermatic cord. The cut surface was white and solid (Fig. 3).

In microscopic findings, edematous, degenerative, and severe inflammatory change with infiltration of red blood cells and eosinophils was observed in the tumor (Fig. 4). Eosinophilic infiltration and thick, fibrous change was also observed at the sheath of the spermatic cord, close to the tumor. No abnormal findings were observed in the seminal duct, artery, vein of the ductus deferens or testis. Vasculitis was not present. Immunostaining for T and B cells revealed no lymphoma or plasmacytoma. The final histopathological diagnosis was eosinophilic funiculitis.

The patient's postoperative course was uneventful, and he was discharged from the hospital twelve days postoperatively. Five days after the operation, the testicular tumor markers such as alpha-fetoprotein and human chorionic gonadotropin were not elevated respectively (3.3 ng/ml; normal range 0.0–9.99 ng/ml, ≤1.0 mIU/ml; normal range 0.0–2.7 mIU/ml). Three days after the operation, the eosinophil count increased to 1462/μL. Twenty-seven days postoperatively, lactate dehydrogenase were still slightly elevated (237 IU/L), the eosinophil count decreased to the normal range and the immunoglobulin E (IgE) level increased to 4610 IU/mL (normal range, ≤173 IU/mL). The levels of other immunoglobulins, anti-nuclear antibody, perinuclear ANCA, cytoplasmic ANCA, rheumatoid factor and other self-directed antibodies were within normal limits. Thirty-two days postoperatively, 18-F-fluorodeoxyglucose positron emission tomography/CT showed that the enlarged intraabdominal lymph nodes that had been seen on preoperative CT were smaller, and no 18-F-

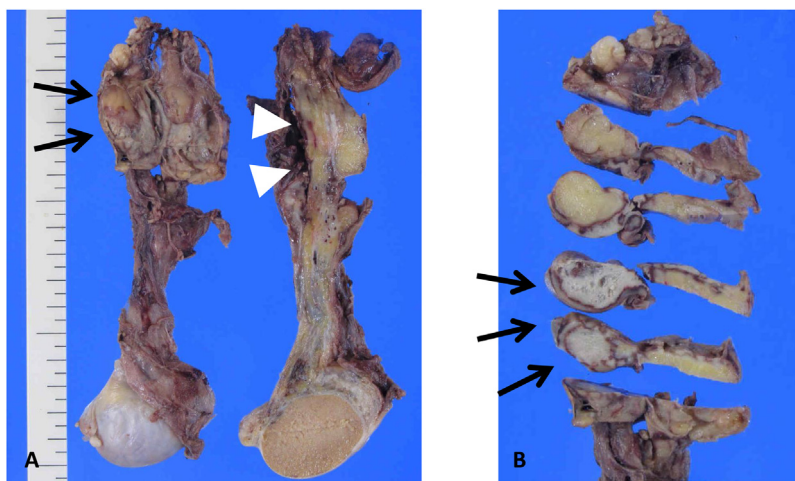


Fig. 3. Gross finding of the specimen. (A) 4 × 3 cm sized tumor (arrow) was located at the proximal of the spermatic cord (arrow head). (B) On the cut section, the tumor showed the white and solid lesion (arrow).

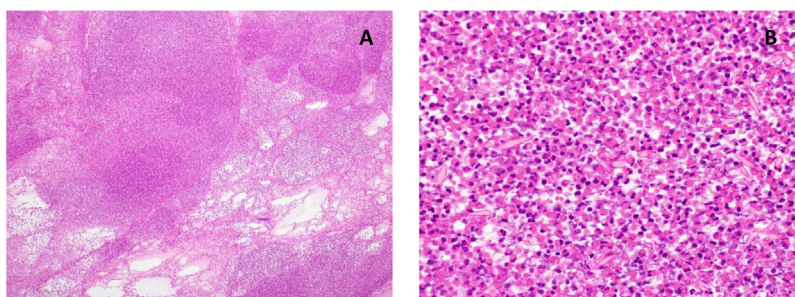


Fig. 4. Microscopic findings. (A) Edematous, degenerative and severe inflammatory change with amount of red blood cells and eosinophils infiltration was observed in the tumor (×40). (B) Significant eosinophils infiltration was observed in the tumor (×400).

fluorodeoxyglucose accumulation was observed in them. At the time of writing, the patient was continuing to undergo periodic follow-up involving blood testing and CT examination by his family doctor.

3. Discussion

Groin hernias are the most common cause of groin masses, which are reportedly present in 84% of cases of groin masses as shown by ultrasonography [1]. Other diseases that cause masses to form in the groin region are divided into benign and malignant tumors. The former are further divided into neoplasms such as spermatic cord lipomas or neurofibromas and non-neoplasms such as infectious tumors or inflammatory hematomas, abscesses, hydroceles, or funiculitis. Most malignant tumors are sarcomas such as liposarcomas, rhabdomyosarcomas, or leiomyosarcomas, and others include metastatic carcinomas, lymphomas, testicular carcinomas, or pseudomyxomaperitonei [2–4].

Benign tumors such as lipomas comprise 70–77% of tumors limited to the spermatic cord and para-testicular tissue, and the most frequent malignant tumors are sarcoma or metastatic carcinoma [2–4]. Testicular carcinoma is the most common solid malignancy affecting males between the ages of 15 and 35, although it accounts for only 1% of all cancers in men [9]. Germ cell tumors account for 95% of testicular cancers. 10–15% of them spread to the spermatic cord or epididymis. Additionally, some reports have described cases of funiculitis caused by bacterial infection or parasitic infestation [10–12]. In our case, elevation of lactate dehydrogenase and retroperitoneal lymphadenopathy such as para-aortic tissue met malignant tumor such as testicular cancer spreading to spermatic

cord, however we didn't suspect the malignancy disease, because no tumor lesions were recognized by ultrasonography in the testis.

We found only two cases of eosinophilic funiculitis in our search of the Japan Medical Literature Database (Igaku Chuo Zasshi) using the key words “eosinophil infiltration,” “funiculitis” or “eosinophil,” and “spermatic cord” [13,14]. When we searched PubMed using the term “eosinophilic funiculitis,” we found no reports except one case already present in the Japan Medical Literature Database.

The most common diseases associated with eosinophilic infiltration are eosinophilic pneumonia and eosinophilic digestive disease. There are few case reports of eosinophilic cholecystitis, cholangitis, pancreatitis, or cystitis [15–19], which are sometimes accompanied by systemic diseases such as hypereosinophilic syndrome or Churg–Strauss syndrome [15,20].

The etiopathogenesis of many diseases associated with eosinophilic infiltration remains obscure. Although smoking is thought to be the cause of some cases of acute eosinophilic pneumonia, the etiology of chronic eosinophilic pneumonia is unclear [5]; therefore, this condition has recently been termed idiopathic eosinophilic pneumonia. An allergic reaction is thought to be one of the causes of eosinophilic digestive disease because eosinophilia and IgE elevation in the peripheral blood are observed, allergic complications such as bronchial asthma are often present, and glucocorticoids are an effective treatment [6]. Possible causes of eosinophilic cholecystitis include medications such as sulfasalazine or herbal medicines, parasite infestations, gallstones, and allergies [16].

Our patient showed no clinical features suggestive of infection and was taking no drugs. He had a history of dermal pruritus, and his child had atopic dermatitis, which is reportedly associated with

eosinophilia. His pruritus might have been due to an allergic reaction, but eosinophilia was not observed on an outpatient blood test. Eosinophilia was observed the day before the operation (1157/ μ L) and 3 and 7 days after the operation (1357–1462/ μ L), and it was temporary. Therefore, it is improbable that an allergic reaction was the cause of this patient's eosinophilic funiculitis. The laboratory data showed no inflammation, so an infectious agent also did not seem to be the cause. No bacteria, parasite eggs, or microorganisms were present in the resected sample, decreasing the likelihood of infectious disease such as parasite infestation (such as schistosomiasis, which is discussed in the existing literature [10]) or bacterial infection. No patients with schistosomiasis have been reported in Japan since 1976 [21], and our patient showed no clinical manifestations indicative of bacterial infection. Based on these results, it is also unlikely that infection was the cause of the eosinophilic funiculitis.

A high IgE concentration was observed after the final diagnosis and was not subsequently rechecked. The levels of other immunoglobulins, anti-nuclear antibody, perinuclear ANCA, cytoplasmic ANCA, rheumatoid factor, and other self-directed antibodies were within normal limits. Based on these results, localized inflammation accompanied by systemic disease such as collagen disease did not seem to contribute to this case of eosinophilic funiculitis.

The other two previously described patients with eosinophilic funiculitis presented with painful groin bulging, fever, and an inflammatory reaction and thus underwent emergent operations for a suspected incarcerated groin hernia [13,14]. In our case, we diagnosed the tumor as an irreducible but not incarcerated hernia because the pain was under control, laboratory data did not worsen, and no ischemic signs were observed by CT. Therefore, we performed elective surgery.

As a malignant tumor was not completely ruled out, dissection of the tumor from the testicular vessels was necessary. However, this might have caused testicular necrosis when vessels were injured, and might result in residual tumor. So we took a decision of high orchiectomy, after obtaining intraoperative informed consent from the patient's family. But it's undeniable fact that high orchiectomy is too much invasive operation as the result that the dissecting tumor is benign. So we could have considered intraoperative histological examination, or stopped the operation, and later, we would do reoperation after fully explanation to the patient himself.

4. Conclusions

If the groin hernia is not typical in that bulge is irreducible but no inflammatory response is observed for a long period of time, as in our case, it is necessary to consider a differential diagnosis. It is important to make more accurate diagnoses by combining detailed medical examination findings, the patient's history, blood test results, and the findings of imaging studies such as ultrasonography, CT, or magnetic resonance imaging. If malignancy is suspected, 18-F-fluorodeoxyglucose positron emission tomography/CT or needle biopsy might be considered preoperatively, and it is important to obtain informed consent about the necessity of intraoperative pathological examination, its limitations for an accurate diagnosis, and the possibility of infertility; the most appropriate operative procedure must then be chosen.

Conflicts of interest

The all authors have no conflicts of interest to declare.

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Ethical approval

Approval to publish this case report was waived by the institution.

Consent

Written informed consent was obtained from the patient for publication of this case report and any accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal.

Authors' contribution

KY and AI contributed to the data collection and drafted the manuscript. AI and HM participated in the surgery. SK provided the pathological suggestion. All authors participated in the daily medical treatment of the patient. SY contributed to the review of the manuscript. All authors read and approved the final manuscript.

Guarantors

Dr. Akashi Ikubo and Dr. Seiji Yunotani.

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