Malignant Mesothelioma of Tunica Vaginalis Testis complicating a hydrocele

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Abstract

Background

Malignant Mesothelioma of Tunica Vaginalis testis (MMTVT) is a very rare tumour which most medical practitioners would not come across in their life time. In view of this most practitioners would be unaware of the characteristic features of the tumour.

Objective: To report a case of malignant mesothelioma of the tunica vaginalis testis with a review of the literature.

Case Report:

A 69-years-old man presented with a left hemi-scrotal mass which was diagnosed clinically and by ultrasound scan to be consistent with left sided hydrocele. He underwent a left sided hydrocelectomy. He developed a further left sided hemi-scrotal oedematous painful scrotal swelling two weeks post-operatively. Ultrasound scan revealed a large left hemi scrotal swelling with septae and echogenic debris compressing both testicles and poor Doppler flow to the left testis. The scrotum was explored and the intra-scrotal collection was evacuated and left orchidectomy as well as excision of the surrounding oedematous tissue was performed. Histological examination of the excised Tunica Vaginalis revealed malignant mesothelioma. Literature review confirmed that MMTV is a rare condition. The Diagnosis must be suspected when a nodule or nodules are found on physical examination or scrotal ultrasound. Immuno-histochemistry is required for confirmation of diagnosis.

Conclusions:

Pre-operative diagnosis of malignant mesothelioma of tunica vaginalis testis is usually missed in the absence of a nodule or nodules on physical examination or scrotal ultrasound scan. Histopathological examination with immunohistochemistry is the usual way of confirming the diagnosis of MMTVT.

Key Words: Malignant; Mesothelioma; Tunica vaginalis; Testis; Hydrocele.

Introduction

Malignant mesothelioma is a diffuse mass that usually spreads widely in the pleural space and it is associated with extensive pleural effusion as well as direct invasion to thoracic structures besides metastatic spread, 1,2,3, Mesothelioma is also known to arise in the peritoneum, pericardium or tunica vaginalis uncommonly, 4,5. Malignant mesothelioma of the tunica vaginalis testis is rare and occurs in 0.3 to 5 % of all malignant mesotheliomas, 6, 7, 8. The tumour could be solid or cystic with yellow or white colour and a relatively firm consistency. Since its first description in 1957 by Barbera and Rubino, 9 only about 90 cases have been reported to our knowledge in the world literature, 10, 11. We report a case of malignant mesothelioma of tunica vaginalis testis in a 69 year old man which was diagnosed post operatively as a surprise with a review of the literature.

Case Report

A 69 year old man was referred by a physician because of a huge swelling of his left hemi-scrotum. The swelling was confirmed by clinical examination and ultrasound scan to be a moderate sized left hydrocele. The ultrasound scan was also reported as showing normal appearances of the left testis and epididymis. No fluid was seen in the right side of the scrotum with normal testes and epididymis. He was therefore listed for left hydrocelectomy which was performed 2 months later.

A Jaboulay’s left hydrocelectomy was performed. During the procedure, it was observed that the hydrocele sac was very thick walled. Therefore, the excised sac wall was submitted for histological examination. The Diagnosis must be suspected when a nodule or nodules are found on clinical examination or ultrasound. Immuno-histochemistry is required for confirmation of diagnosis.

A 69 year old man was referred by a physician because of a huge swelling of his left hemi-scrotum. The swelling was confirmed by clinical examination and ultrasound scan to be a moderate sized left hydrocele. The ultrasound scan was also reported as showing normal appearances of the left testis and epididymis. No fluid was seen in the right side of the scrotum with normal testes and epididymis. He was therefore listed for left hydrocelectomy which was performed 2 months later. A Jaboulay’s left hydrocelectomy was performed. During the procedure, it was observed that the hydrocele sac was very thick walled. Therefore, the excised sac wall was submitted for histological examination. A redivac drain was inserted because of oozing from the thickened wall of the hydrocele and the drain was removed after 3 days when the drainage was noted to be minimal. He was put on antibiotics. He was discharged home after 4 days.

Two weeks post-operatively, the patient was seen in the clinic and readmitted because his left hemi-scrotum was swollen, sore and oedematous. Ultrasound scan of the scrotum which was performed on admission was reported as follows: (a) A big collection in the left hemi-scrotum with septae and echogenic debris compressing both testicles. (b) The
left testicle showed a poor or absent doppler flow and may be compromised. (c) The right testicle appeared within normal limits. (c) There is oedema of the scrotal wall. (d) The left testicle may be compromised by the huge post operative haematoma. In view of these findings the scrotum was explored and the intra-scrotal collection was evacuated and left orchidectomy as well as excision of the surrounding oedematous tissue was performed. The cavity within the scrotum was initially packed and the pack was subsequently removed before the patient was discharged to continue having wound dressing at home.

The patient was reviewed in the clinic about 7 weeks after the initial operation and it was observed that even though the inflammation in the left hemi-scrotum was settling there was an indurated mass surrounding a sinus in the left hemi-scrotum. At this stage the final histology report of the excised tunica (the wall of the hydrocele sac) was available. The provisional report indicated that the histology revealed: (a) Tumours which showed a complex micro-papillary and tubular pattern. (b) There was moderate nuclear pleomorphism. The appearances were suggestive of mesothelioma (See figures 1 and 2). The delay in obtaining the report was due to the fact that further examination of the specimen and second opinion had to be obtained. The supplementary report confirmed that the patient had malignant mesothelioma of tunica vaginalis of testis. Immunohistological staining of the tumour for calretinin which is a mesothelial tumour marker was positive (See figure 3).

In view of the histological findings a CT scan of thorax, abdomen and pelvis with contrast was requested and the case was referred to the Multi-Disciplinary Team Meeting. The CT scan findings were as follows:

A few shotty mediastinal lymph nodes, some of which contain calcium. There is a hiatus hernia with most of the stomach being situated within the chest. Normal appearances of the central airways and lung parenchyma with no evidence of pulmonary metastases.

Below the diaphragm, para-aortic lymphadenopathy is noted measuring 18 mm in short axis diameter to the right and 33 mm in short axis diameter to the left.

Normal appearances of the liver, pancreas and spleen with no evidence of metastases.

Normal appearances of the left kidney. The right kidney contains a solitary simple cyst. Some degenerate changes are noted in the lumbar spine, but there is no evidence of bony metastases.

Normal appearances of the bones.

The CT scan report was therefore, summarized as:

(a) Both right and left sided para-aortic lymphadenopathy was noted in the abdomen but there are no pelvic lymph nodes.

(b) No evidence of visceral metastases.

(c) Normal appearances of the bones.

It was decided at the Multi-Disciplinary meeting that histology of the specimen excised from the scrotum should be further reviewed at the Regional Oncology Centre and the patient should also be seen at the Regional Oncology Centre. A review of the specimen at the Regional Oncology Centre confirmed that Malignant Mesothelioma of Tunica Vaginalis of testis had complicated the hydrocele. The patient was seen by Oncologists at the Regional Oncology Centre who had explained that radiotherapy and chemotherapy have no place in the management of malignant mesothelioma of tunica vaginalis testis because if at all the success rate is just about 15%. The Oncologists explained to him that:

1) A repeat CT scan had suggested possibly some residual swelling.

2) The Oncologists will adopt a wait and see or active surveillance approach.

3) A CT scan will be performed later (soon).

4) If there is evidence of a further growth then he would be considered for further surgical resection after discussions.

Discussion

Mesothelial tumours can arise from any tissue where mesothelial membrane exists, and as the tunica vaginalis is derived embryologically from outpouching of the peritoneal cavity, it is rarely the site of primary mesothelioma. The development of malignant mesothelioma has been proven to be related to exposure to asbestos or different asbestos –containing materials (34.2% of patients) 12; 13. This patient did not have any exposure to asbestos to our knowledge. A causal association to prior radiotherapy has not been established14. Trauma and inguinal herniotomy are also considered predisposing factors 15; 16. Malignant mesothelioma of tunica vaginalis testis is diagnosed most frequently (56.3%) at the time of hydrocelectomy for presumed benign hydrocele17. Most other cases are diagnosed preoperatively as testicular tumours (33%) and orchietomy is performed18. The tumour presents most commonly between the 5th and 7th decades (mean 53.5 years) but it has been reported in patients as young as 7 years13. However, 20% of previous 76 cases reported affected the first 3 decades, and the prognosis is better for this group of patients and 10% of the patients were younger than 25 years 12; 19. Malignant mesothelioma of tunica vaginalis testis presents as
hydrocele or a firm mass (tumour mass) may be palpated often in association with a hydrocele but unfortunately the preoperative diagnosis is made in only 3% of cases12; 19. Mesotheliomas present variable histological patterns including: (a) epithelial, (b) fibrosis, (c) and mixed types (biphasic - composed of epithelial and sarcomatoid components) 1; 2. Variations in the histological types of mesotheliomas are said to be due to tumour location. Pleural mesotheliomas have been stated to be predominantly fibrous, whereas, mesotheliomas of the peritoneum and tunica vaginalis have been noted to be epithelial3. Mesothelial cells develop as cuboidal, columnar or papillary structures that resemble adenocarcinoma. When the ensuing features are present then the diagnosis favours mesothelioma as opposed to adenocarcinoma:

(a) Lack of staining for carcino-embryonic antigen (CEA)
(b) Strong staining for vimentin and presence of epithelial glycoprotein 4; 5.
Reactive mesothelial hyperplasia secondary to hydrocele or an underlying inflammatory process is ruled out by presence of atypism besides lack of a history of those predisposing factors mentioned above 5.

The criteria for malignancy are nuclear pleomorphism, mitotic activity and stromal invasion. Positive immunohistochemical staining antibodies of cytokeratin and vimentin, negative immunostaining for carcino-embryonic antigen and Leu-M1 usually establish the diagnosis20. These methods are used to differentiate mesothelioma from adenocarcinoma metastasis. Positive staining for factor viii related antigen is also a useful way of excluding histiocytoid hemangioma17.

The epithelial type mesothelioma is reported to account for 75% of tumours (In 75% of cases, microscopic examination reveals epithelial mesothelioma) and its first differential diagnosis is florid mesothelial hyperplasia. Mesothelial hyperplasia is a benign lesion, and the tumour mass formation excludes malignant mesothelioma. The clinical profile of carcinoma of the rete testis overlaps with malignant mesothelioma, due to the fact both tumours have papillary and tubular patterns19. However, the location of the tumour within the rete testis, and evidence of continuity with the rete testis as well as immunohistochemical positivity for carcinoembryonic antigen are important in distinguishing the two lesions.

Even though pathophysiologic mechanism for the development of malignant mesothelioma related to asbestos exposure is not clear, Karunharan T suggested that direct effect of asbestos fibres on the tunica vaginalis or the deposition of asbestos fibres within the serosa causing chronic irritation of the tissue could be another reason for the development of mesothelioma21. Guardal M and Erol A, 22 believe that chronic hydrocele may be the cause of malignant mesothelioma by chronic irritation.

Pre operative diagnosis of malignant mesothelioma is usually missed in the absence of nodules on physical examination or ultrasonographic examination. In our case, the diagnosis was not obvious preoperatively on clinical examination or on ultrasound scan. Even the intraoperative finding of thickening of the tunica was not sufficient to indicate the diagnosis of malignant mesothelioma. Because ultrasonography, computed tomography or MRI can usually depict clearly the vaginalis testis cavity of the hydrocele, the characteristic gross appearance of the mesothelioma could sometimes be delineated by these imaging techniques. Four case reports have described the ultrasonographic features23; 24; 25; 26. Tyagi and co-workers23, reported discrete echogenic nodules up to 1 cm in diameter studding the scrotal sac lining. Fields and co-workers24, described bilaminar fronds extending from the scrotal lining into the hydrocele. Fields and co-workers also stated that it is difficult to diagnose mesothelioma from cytology of aspiration fluid of the testicular hydrocele. In our case there was no suspicion of mesothelioma and hydrocele fluid was not sent for cytology.

Fujisaki and co-workers26, used ultrasound and MRI scan to show nodular or papillary projection of tumours into the hydrocele cavity. In their report, the ultrasound showed a slightly hypoechoic nodular mass attached to the parietal vaginal layer and ultrasound taken two years later showed exacerbated thickening of the scrotal wall and a newly developed cystic component. In the same report, T1-weighted magnetic resonance imaging revealed nodular masses studding on the hydrocele sac, and two years later T1-weighted magnetic resonance imaging showed multiple and non-homogenous cystic components.

It has been suggested that cytology may be diagnostic in suspicious cases27. Standard guidelines regarding the extent of resection have not been established, but early aggressive resection would appear to be the main modality of treatment for malignant mesothelioma of the tunica vaginalis testis 12; 13; 15. It has been stated that these patients are best treated by radical inguinal orchiectomy in all cases, and in cases where the diagnosis is made after hydrocelectomy or exploration.
of the scrotum then hemiscrotectomy should be performed 12; 13; 15. The role of inguinal, pelvic and retroperitoneal lymph node dissection in malignant mesothelioma of tunica vaginalis testis is controversial. Gupta and co-workers as well as Jones and co-workers recommend ipsilateral inguinal lymphadenectomy 13; 15. It has been recommended that retroperitoneal lymphadenectomy should be performed if evidence of nodal disease is present 13; 15. Positive lymph nodes were in 10 of 74 patients (3 inguinal, 2 pelvic and 5 retroperitoneal) at the time of diagnosis in the review of Plas and co-workers 12. Surgical excision of local recurrences is the best option of treatment currently available. Mesothelioma usually has an aggressive behaviour if it is disseminated 28. Mortality rate was reported to be 53% with a mean follow-up of two years 15. Plas and co-workers reviewed 73 malignant mesotheliomas of tunica vaginalis reported over 30 years and made the following observations:

- The median survival was 23 months
- Recurrence occurred in 52.5% of cases and it decreased survival to 14 months
- About 20% of cases occurred in patients in the first 3 decades and the prognosis is better for this group of patients 12.

Radiotherapy and various regimes of chemotherapy were given without achieving any remission. However, Black and co-workers observed partial remission in one patient who was treated with gemcitabine and cisplatin 18.

Sixty percent of recurrences occur within 2 years of follow-up, and local recurrence was reported in 23.7% of patients, with scrotal involvement in 10% of the cases. The involvement of inguinal or iliac lymph nodes occurs in 3 to 5% of the cases and the role of their dissection is the subject of discussion. Visceral metastases are rare and the involvement of liver and lung is reported in 4% and 10% respectively. Cutaneous invasion and bony metastasis may occur.

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References

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Illustrations

Illustration 1

Figure 1 shows malignant mesothelioma of tunica vaginalis at 40X magnification
Illustration 2

Figure 2 shows malignant mesothelioma of tunica vaginalis at 200X magnification
Illustration 3

Figure 3. Shows tumour positively stained for calretinin which is a mesothelial tumour marker
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