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PRESERVATION OF BLADDER IN RECURRENT BLADDER CANCER

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ABSTRACT

The urachus carcinoma is an extremely rare tumor that accounts for less than 1% of all bladder cancers. Because of the rarity of presentation and lack of good volume cases, there are no universally accepted diagnostic criteria available to diagnose these cases. Management of urachal adenocarcinoma or rather bladder adenocarcinoma is unclear with lack of universal staging criteria and management protocol. We report a case of a 52-year-old lady, who was diagnosed with adenocarcinoma bladder. After thorough search of the literature, we have discussed the various management options available for managing such cases.

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INTRODUCTION

Urachal cancers are rare and aggressive cancers of the bladder which were originally described by Hue and Jacquin in 1863. The first extensive description of this entity was made by Begg in 1931 (Paner, 2016 and Begg, 1930). Adenocarcinoma of bladder accounts for less than 1% of all bladder malignancies. The urachus carcinoma accounts for <0.3% of all bladder cancers (Bruins, 2012 and Pinthus, 2006). It is a malignant epithelial neoplasm arising from urachal remnants. Ninety percent of them are adenocarcinomas (Szarvas, 2016), believed to evolve from intestinal metaplasia of the epithelial component (Paner, 2016). Nonglandular neoplasms can be urothelial, squamous cells, neuroendocrine, and mixed type (Moch, 2016). To date, because of its rarity, there is some inconsistency and no consensus in the literature about the nomenclature, the diagnostic criteria, the staging system to use, and the best therapeutic options.

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We report a case of 52-year-old woman with an urachal carcinoma and discuss briefly about it with reference to literature.

Case Report

Our patient was a 52 year old lady, who was a diagnosed case of adenocarcinoma urinary bladder and underwent partial cystectomy at a hospital before 9 months. She presented to us with complaints of abdominal pain with increased frequency of micturition for one month duration. On examining her we found a mass of size 4x4 cm present over the hypogastrium, which was even present after micturition. Routine investigations were within normal limits. Then we proceeded with CECT whole abdomen and pelvis which revealed a 4.2 x 3 cm mass seen at dome and anterior wall of the bladder with perivesicular extension and mild tethering of rectus sheath [? urachal remnant origin]. Metastatic workup revealed no evidence of metastasis. She was planned for a diagnostic cystoscopy, which showed a 1.5 cm proliferative growth present in the dome of the bladder at the site of urachal attachment. Cystoscopy biopsy was done and the specimen

was sent for histopathological examination (HPE). HPE showed mucin secreting adenocarcinoma of bladder with invasion of muscular layer. This belonged to stage III B in the Sheldon system and stage II in the Mayo system. Patient was then planned for partial cystectomy with urachus excision and omphalectomy. Intra operatively we found a nodular mass of size 4 x 3 cm involving the bladder dome extending into the prevesical space and also involves the rectus muscle. The mass was excised along with the urachus and the dome of the bladder with a 2 cm margin. Part of rectus muscle and umbilicus were also included with the specimen. Nodal dissection was not done. Bladder was closed in two layers. Abdomen was closed in layers. Post op histopathology report showed adenocarcinoma of urachal remnant of size 3.5 *1.5 cm and all resected margins were free of tumour. Patient was kept on followup with repeated physical examination, imaging and diagnostic cystoscopy. Two year follow up revealed no evidence of recurrence or metastasis.

DISCUSSION

The urachus is a vestigial structure that connects the bladder to the allantois during early embryonic development. After birth it becomes a fibrous cord known as the median umbilical ligament. If remnants of the allantois remain within the ligament, they may develop into cysts and epithelial neoplasms. Urachal remnants have been identified in one third of cases in post mortem studies; in the dome and anterior wall commonly bladder (Begg, 1982 and Schubert, 1982). The urachus has intramucosal, intramuscular and supravesical segments. It contains three distinct tissue layers: an epithelial canal lined by urothelium, submucosal connective tissues and an outer layer of smooth muscle. Urachal neoplasms can arise in any of these layers, and can be epithelial or mesenchymal. Similar to urothelium at other sites, the epithelium often demonstrates focal glandular metaplasia, and this provides a morphologic basis for the development of intestinal-type tumors (Begg, 1930 and Schubert, 1982). In the 1950s Wheeler and Hill proposed the initially accepted diagnostic criteria for the urachus carcinoma. The criteria has undergone modifications and is still controversial; even so, most investigators accepted the proposed criteria by Sheldon *et al*. (1982) and Mostofi *et al*. (Mostofi, 1955) that were (a) tumor in the dome of the bladder, (b) absence of cystitis cystica and cystitis glandularis, (c) predominant invasion of the muscularis or deeper tissues with a sharp demarcation between the tumor and surface bladder urothelium that is free of glandular or polypoid proliferation, (d) presence of urachal remnants within the tumor, (e) extension of tumor into the bladder wall with involvement of the space of Retzius, anterior abdominal wall, or umbilicus, and (f) no evidence of a primary neoplasm elsewhere. However, these criteria were considered somewhat restrictive by some studies (Dhillon, 2015; Gopalan, 2009 and Johnson, 1985).

A new somewhat broader set of criteria adapted from Gopalan *et al*. was published in the 2016 World Health Organization (WHO) blue book for the diagnosis of urachal adenocarcinoma which are (a) location of the tumor in the bladder dome and/or anterior wall, (b) epicenter of carcinoma in the bladder wall, (c) absence of widespread cystitis cystica and/or cystitis glandularis beyond the dome and anterior wall, and (d) absence of a known primary tumor elsewhere. The case presented here illustrated the following features: the tumor was located in the anterior wall and in the bladder dome, the epicenter of the tumor was in the bladder wall, there was absence of cystitis cystica or cystitis glandularis, and the investigations carried out did not reveal any primary tumor elsewhere, thereby fulfilling all the WHO criteria for the diagnosis of urachal adenocarcinoma. It showed also a sharp demarcation between the tumor and surface bladder urothelium that was free of glandular and polypoid proliferation. And finally the presence of urachal remnant explained by the tumor extent was not documented. This feature is helpful for the diagnosis but its absence does not preclude the urachal origin. The most frequent initial symptom is hematuria but mucinuria (mucin in the urine), local pain or swelling, recurrent local or urinary tract infections, abdominal lump and umbilical discharge can be seen (Paner, 2016 and Szarvas, 2016). There are several staging systems for urachal carcinomas. The most widely accepted one is the staging proposed by Sheldon *et al*. (Table 1) but their relevance still needs validation by larger series.

Staging of urachal adenocarcinoma (Sheldon's system).

Stage	Characteristics	
I	Tumor limited to vesico-urachal mucosa	
II	Tumor invaded the submucous or muscularis but confined to the urachus	
III	Invasion beyond the urachus	A. Extension to bladder B. Extension to abdominal wall C. Extension to peritoneum D. Extension to local viscera
IV	Metástasis	A. Metastasis to regional lymph nodes B. Distant metastasis

The recommended treatment for nonmetastatic cases is surgery. Partial or radical cystectomy has similar oncologic results (Szarvas, 2016). An en bloc resection of the urachal ligament and umbilicus is recommended for patients who have surgically resectable disease and either a complete or partial cystectomy ensuring negative margins. This is crucial because urachal tumours can occur anywhere along the urachus, including at the umbilicus (7%). If the urachus is transected during surgery, spillage of the tumour containing fluid into the peritoneal cavity can increase the risk of relapse (Ashley, 2006 and Sheldon, 1984). The effective role of neoadjuvant or adjuvant chemotherapy is still to be proven. However there are some reports of metastatic cases, with response to FOLFOX chemotherapeutic regimen (Paner, 2016).

Conclusion

Urachal adenocarcinoma is an uncommon malignancy that often presents late because of its location and relatively non-specific symptoms. This probably results in its poor prognosis. Adequate primary surgery and close follow up is the treatment of choice. Complete urachectomy and umbilectomy were significant predictors of survival and recurrence. Local recurrence may be due to seeding within the distal urothelial tract, particularly in tumors with a configuration that is polypoid and which open into the bladder cavity. The type of surgery performed may have an effect on local recurrence despite negative margins of resection. Bladder preserving surgery can still play a role in the management of adenocarcinoma of bladder.

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