Primary Appendiceal Adenocarcinoma Masquerading as Primary Bladder Tumor: a Case Report and Review of Literatures

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Abstract Adenocarcinoma of the appendix invading the urinary bladder is very rare. We describe such a case in a 42-year-old man and review the relevant literatures. In the present case, although ultrasonography and computed tomography scan revealed a mass in the bladder or in the pelvic and biopsies by cystoscopy showed an adenocarcinoma suspicious for primary of the bladder, its real origin was hard to distinguish. An open exploratory surgery was performed with subsequent resection of appendix and partial bladder. Postoperative histopathological examinations revealed the appendiceal adenocarcinoma infiltrating the bladder wall. The patient refused the right hemicolectomy and received 3 cycles of adjuvant radiotherapy. However, tumor recurrence was found in the bladder 6 months after surgery, and radical cystectomy was eventually performed.

Keywords Adenocarcinoma · Appendiceal neoplasms · Urinary bladder neoplasms

Introduction

Primary appendiceal adenocarcinoma is a very rare condition that constitutes 0.4–1.0 % of all malignancies of gastrointestinal origin [1]. Adenocarcinoma of the appendix invading the urinary bladder is even rarer with only isolated cases being

reported. Here, we describe a case of appendiceal adenocarcinoma in a patient who presented with only irritative urinary symptoms and was initially diagnosed as having a bladder tumor.

Case Report

A 42-year-old man, who had undergone computed tomography (CT) scan and pathological examination at another hospital, presented at our outpatient clinic with a 1-year history of micturition pain and urinary frequency. The non-enhanced CT showed a solid mass in the bladder, adhering to the adjacent intestines, and pathological examinations by transurethral cystoscopy revealed an adenocarcinoma suspicious for primary of the bladder. He had no complaints of any symptoms referable to the gastrointestinal tract except a recent onset of anus bulge discomfort.

Physical examination was negative, but our enhanced CT scan revealed a pelvic mass at the level of the right anterior bladder wall, measuring 4.8×4.1 cm, suspicious for bladder or bowel or other origin (Fig. 1). Barium enema was subsequently performed but showed no positive findings. On laboratory investigation, micro-hematuria was observed, and the patient's carcinoembryonic antigen level was 5.15 ng/ml, which was slightly elevated, but other biochemical markers were normal.

At the open exploratory surgery, an abdominal mass localizing between the bladder and the cecum was found, measuring 4×5 cm, adjoining the appendix and deeply infiltrating the bladder wall. After consulting with general surgeons and having a confirmation of no other tumors being observed, we resected the whole mass and subsequently performed an appendectomy and partial cystectomy limited to the invaded bladder wall. Postoperative histopathological examinations revealed a low to

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Fig. 1 CT scan image of a pelvic mass at the level of the right anterior bladder wall, measuring 4.8×4.1 cm

moderately differentiated adenocarcinoma with mucinous components, originating from the appendix (Fig. 2), infiltrating the bladder wall and extending to its submucosa. The patient refused right hemicolectomy and was discharged 7 days after operation. Cycles of adjuvant radiotherapy were scheduled for him in our department of oncology. However, 6 months after the surgery, tumor reoccurred in the bladder, and radical cystectomy was eventually performed. The patient is now stable with normal carcinoembryonic antigen level (2.65 ng/ml) and is under close surveillance and long-term follow-up.

Discussion

Malignant tumors of the appendix invading urinary bladder are extremely uncommon. As lack of typical symptoms and

Fig. 2 Postoperative histopathological examination image of a low to moderately differentiated adenocarcinoma with mucinous components (HE-100)

representative imaging presentations, correct diagnosis is usually difficult to be obtained preoperatively. Most patients present with only urinary symptoms, such as hematuria, micturition pain, urinary frequency, etc., but fecaluria may strongly suggest an enterovesical fistula formation, which is usually due to untreated acute appendicitis or appendiceal carcinoma [2]. Of the literature reviewed, none of the patients was diagnosed to have appendicitis preoperatively, which may imply that the patients with longer duration of both abdominal and urinary symptoms should be suspected of a more malignant disease such as appendiceal carcinoma [3].

On laboratory tests, the elevated carcinoembryonic antigen level is reported in some cases [2, 4] and may hint a tumor of gastrointestinal origin. On radiographic examinations, enhanced CT scan is valuable and necessary for us to suspect the previous conclusion. Barium enema sometimes reveals extrinsic compression at ileocecum or formation of an enterovesical fistula and helps to a better diagnosis [4], but we attained no positive findings.

Immunohistochemical approaches are mentioned to determine the origin of the malignancy [3]. The panel of cytokeratin (CK)7, CK20, thrombomodulin (TM), and β -catenin has been recommended to discriminate between primary bladder adenocarcinoma and secondary colorectal adenocarcinoma. The profile of CK7–/CK20+/TM–/ β -catenin+ is usually observed in secondary colorectal adenocarcinoma, while CK7+/CK20+/TM+/ β -catenin– may be suggestive of primary bladder adenocarcinoma [5]. The study of Chu [6] analyzing 175 cases demonstrated that the immunohistochemical profiles of primary bladder mucinous adenocarcinomas (MAs) were similar to those of appendiceal MA: positive for CK7, CK20, CDX2, and mucin-2 (MUC-2) and negative for MUC-1, MUC-6, and β -catenin, while colonic MA showed positive results for CK20, CDX2, MUC-2, and β -catenin and negative for CK7, MUC-1, and MUC-6. Thus, it seems relatively easier to distinguish the bladder adenocarcinoma from colonic or rectal adenocarcinoma rather than that from the appendix.

Although there was no evidence of lymph node metastasis and surgical margins were all negative, tumor recurrence was still observed in the bladder 6 months after surgery, and radical cystectomy was eventually performed. The reported overall 5-year survival of appendiceal adenocarcinomas was 46.2 %, but in subgroups, the data was 47.9 % adenocarcinoma, 47.7 % mucinous adenocarcinoma, 59.0 % mucinous cystadenocarcinoma, 20.3 % signet ring cell carcinoma, and 1.7 % lymphomas, respectively [7]. Surgical approaches also influence the prognosis. The study by Nitecki et al. suggested that the 5-year survival rate was superior after right hemicolectomy versus appendectomy alone (68 vs. 20 %) [8]. As of now, our patient appears fine without any symptoms or recurrence of tumors

6 months after the eventual cystectomy, and he is still under close and long-term follow-up.

Conclusion

Primary appendiceal adenocarcinoma masquerading as a bladder tumor is exceedingly rare. When encountering cases of urinary bladder neoplasm of indistinct origin, we should consider the possibility of secondary tumor from the appendix. As no standard treatment was established for such rare conditions, according to our experience and literature review, we suggest an enhanced CT scan and transurethral biopsies combined with a certain immunohistochemical panel as effective diagnostic methods and recommend appendectomy, right hemicolectomy, and cystectomy as appropriate procedures for such a patient.

Conflict of Interest We declare that none of the authors have any financial and personal relationships with other people or organizations that can inappropriately influence the quality of the work presented in this manuscript.

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