Case Report

A case of primary adenocarcinoma of Meckel's diverticulum presenting as ovarian mass and literature review

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ABSTRACT

Meckel's diverticulum (MD) is the most common congenital anomaly affecting the gastrointestinal tract. It is located on the anti-mesenteric border of the small intestine and occurs due to incomplete obliteration of the omphalomesenteric duct during embryogenesis. Tumors, particularly cancers, are rare complications of MD. Carcinoids are the most common malignancies in this site. Adenocarcinomas are extremely rare and carry a poor prognosis. Here, we present the case of adenocarcinoma of MD presenting as Krukenberg tumor in a 42-year-old lady and causing a diagnostic dilemma. Pelviabdominal examination revealed large bilateral adnexal masses along with ascites and computed tomography-guided fine-needle aspiration cytology from the right adnexal mass was suggestive of adenocarcinoma. At exploratory laparotomy, a MD was found with hard intramural growth palpable at the tip of MD adnexal deposits and omental caking. Immunohistochemistry revealed CK 7- and CK 20+ve and also CDX2 positive. The rarity of presentation of bilateral adnexal mass due to primary adenocarcinoma of MD urges us to report this case.

Key words: Adenocarcinoma, Krukenberg tumor, Meckel's diverticulum, Ovarian tumors

Adenocarcinoma of Meckel's diverticulum (MD) is very rare with only 30 such cases being reported in the English literature [1]. It is even rarer for these patients to present with a Krukenberg tumor [2].

We hereby present a case where a female patient presented with pelvic mass mimicking primary ovarian cancer. After further investigations and during surgery, it was found to be a case of primary adenocarcinoma of MD with Krukenberg tumor. Carcinoma ovary is a commonly encountered problem in any oncology center but adenocarcinoma of MD mimicking carcinoma ovary is very rare and should be considered as a differential diagnosis. Furthermore, Krukenberg tumor from MD was only reported once previously which indicates the rarity and importance of this case report.

CASE REPORT

A 42-year-old premenopausal lady, with no previous major medical illness, hospital admission, or any previous surgical intervention was admitted at our center with complaints of lower abdominal pain and heaviness for 6 months.

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On clinical examination, there was no obvious pallor or icterus and performance status was ECOG 2 (Eastern Cooperative Oncology Group). Pelviabdominal examination revealed large bilateral adnexal masses along with ascites.

Computed tomography (CT) scan chest was normal but that of abdomen and pelvis revealed oval cystic septated bilateral adnexal masses, indistinct uterine outline with retroperitoneal lymphadenopathy, peritoneal caking, and mesenteric nodules. Ovaries were not visualized separately.

Investigations revealed CA-125 levels to be moderately raised at 220 U/ml and carcinoembryonic antigen at 3.8 ng/ml. Ascitic fluid stained positive for malignant cells and CT-guided fine-needle aspiration cytology (FNAC) from the right adnexal mass was suggestive of adenocarcinoma with predominantly papillary features. Upper gastrointestinal endoscopy and colonoscopy were normal. At a multidisciplinary meeting, a possibility of bilateral primary epithelial ovarian malignancy was strongly considered.

Surgical exploration was advised with an aim for cytoreductive surgery. At exploratory laparotomy, a MD was found with hard intramural growth palpable at the tip of MD with extensive serosal, mesenteric, and adnexal deposits and omental caking (Fig. 1).

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FNAC was done from MD and the omentum was biopsied and peritoneal washings were sent for cytological examination. Resection of MD was not felt to be justified in presence of extensive seedlings. The omental biopsy later showed the features of a metastatic moderately differentiated adenocarcinoma. FNAC from the MD showed features of adenocarcinoma. Immunohistochemistry revealed CK 7- and CK 20+ve and also CDX2 positive, suggestive of gastrointestinal origin.

The post-operative course of the patient was uneventful. Considering her extensive metastatic disease and predicted poor response to chemotherapy, after discussion in the multidisciplinary tumor board in presence of next to kin and relatives of the patient, only supportive care had been planned and hence she was transferred to the palliative care department. The patient expired 4½ months later with good symptom control.

DISCUSSION

Although Fabricius Hildamus in 1598 and Lavater in 1671 described diverticular projections from the ileum, it was Johann Meckel, who in a paper in 1809 identified it as a remnant of the vitelline duct that normally connects the yolk sac to the primitive gut [3]. It is a true diverticulum, having all the layers of the intestine. Heterotopic tissue such as gastric, pancreatic, or colonic tissue may be found in the MD.

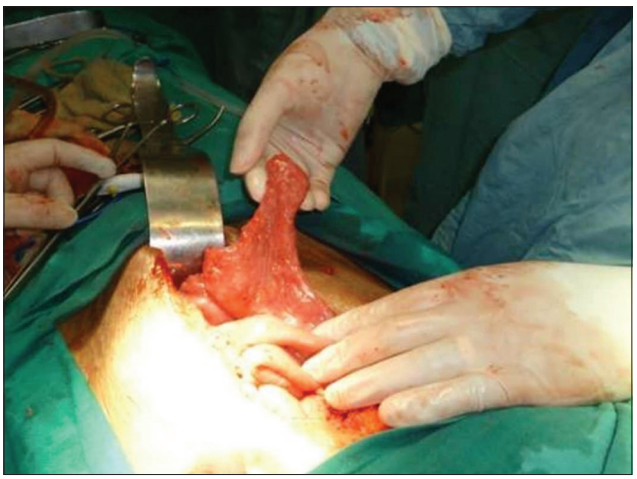

Figure 1: Operative photograph showing intramural mass in the tip of Meckel's diverticulum and extensive peritoneal deposits

In a large population-based study, spanning 42 years, the lifetime risk of complications has been reported as 6.4% which includes hemorrhage (commoner in children), obstruction (commoner in adults), diverticulitis and/or perforation, ulceration, and Littre's hernia [4]. Primary malignancy arising out of MD is not common. In a recent analysis of the SEER database from 1973 to 2006, cancers in MD were found in 163 cases, 77% of which were carcinoids [5,6]. Adenocarcinoma arising from MD is thus extremely rare. Johnson in 1973 reported the total number in the world literature as 18 [7], and recently, Kusumoto *et al*. reported 30 cases [1]. A review of the literature of cases of adenocarcinoma of MD is shown in Table 1 [8-14].

Most of the adenocarcinoma is thought to arise from heterotopic mucosa, and as such, gastric and intestinal types predominate, though another type like pancreas-type adenocarcinoma has been reported [8]. The common modes of presentation are abdominal pain and obstruction and most are detected in a metastatic setting [9]. Treatment of MD adenocarcinoma in the resectable stage includes resection and anastomosis of the small intestine with dissection of regional nodes. The outcome is generally poor [10].

Krukenberg tumors are most often discovered in young women usually in their fifth decade of life. Abdominal pain and distension are the most common presenting complaints. It may be associated with virilization and ascites. The lymphatic system is the most probable route of metastasis in a Krukenberg tumor, but cases without lymphatic involvement have also been reported suggesting peritoneal seeding. Krukenberg tumors are bilateral in most of the cases. The ovaries are usually solid with smooth capsular surfaces and asymmetrically enlarged. Microscopically, the tumor characteristically reveals mucin-laden signet-ring cells within the ovarian stroma. Immunohistochemistry imparts mostly to differentiate between primary ovarian malignancy and colonic tumor metastasizing to the ovary. The mortality of the Krukenberg tumor is significantly high and the prognosis

Table 1: Review of the literature of cases of adenocarcinoma of Meckel's diverticulum

Author	No. of cases	Metastasis	Treatment	Outcome
Kusumoto *et al*. [1]	30			
Sakpal *et al*. [2]	1	Yes, Krukenberg	Surgery	Not reported
Koh *et al*. [8]	1	Yes, peritoneum	Palliative resection	Rapid progression, death at 6 weeks
Muto *et al*. [9]	1	No	Surgery→CT	Recurrence at 6 months
Lippe *et al*. [10]	1	No	Surgery	Recurrence at 1 month; no response to CT
Parente *et al*. [11]	1	Yes	Palliative resection + CT	4-month follow-up
Rieber *et al*. [12]	1	Synchronous gastric cancer, non-metastatic	Synchronous resection	No recurrence at 1 year
Martín *et al*. [13]	1	Yes, mesenteric lymph nodes	Surgery	Not reported
Lin *et al*. [14]	1	No	Surgery	No recurrence at 6 months
Present case	1	Yes, peritoneal, Krukenberg	None	Died within 4 months
Total	39			

CT: Computed tomography

is worse. Surgical resection is considered to be fruitless if the primary tumor remains unknown or residual disease is present after the intervention. Adjuvant chemotherapy appears to have no significant effect on prognosis.

CONCLUSION

The present case deserves mention due to various reasons. First, adenocarcinoma of MD is rare. Second, adenocarcinoma of the MD presenting as Krukenberg tumor is even rarer. We could find only one case reported in the English literature of adenocarcinoma of MD presenting as primary tubo-ovarian mass (Krukenberg tumor).

AUTHORS' CONTRIBUTIONS

Gupta Arnab and Chakraborty Biman were the primary surgeons. Singh Inderdeep was assisting surgeon. Gupta Arnab has written the manuscript and it was reviewed by Singh Inderdeep and Haldar Sudip. All authors read and approved the final manuscript.

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