Letter to the Editor

A rare case of gallbladder cancer in a young patient with horseshoe kidney and Hashimoto's thyroiditis

Durmuş Ali Çetin, Ebubekir Gündeş, Hüseyin Çiyiltepe, Ulaş Aday, Emre Bozdağ, Aziz Serkan Senger, Orhan Uzun, Kamuran Cumhur Değer

Gastroenterological Surgery Department, Kartal Koşuyolu High Speciality and Training Hospital, Istanbul, Turkey

Submitted: 14 December 2016 **Accepted:** 24 December 2016

Arch Med Sci Civil Dis 2016; 1: e139–e140 DOI: 10.5114/amscd.2016.65008 Copyright © 2016 Termedia & Banach

Gallbladder cancer is the fifth most common cancer involving the gastrointestinal tract, but it is the most common malignancy of the biliary tract, accounting for 80–95% of biliary tract cancers [1, 2]. Gallbladder cancer is relatively infrequent, particularly in European countries and America, apart from than regions such as Chile, Poland, and Japan, where the disease is seen more frequently. Gallbladder cancer rates tend to increase with advancing age. The median age was 67 years in a Memorial Sloan-Kettering report of 435 gallbladder cancer patients [3].

In this case we present a young patient with a triad of findings that our patient harbors.

The 26-year-old male patient (M.Ç/Protocol no: 497) was admitted to our hospital with a complaint of abdominal pain for one month. His grandfather had died because of lung cancer. He had an episode of meningitis in childhood, but there was no obvious neurological deficit. There was no pathological finding on physical examination. Horseshoe kidney anomaly, hydropic gallbladder, asymmetrical posterior wall thickness enlargement and a 22 × 22 mm image on the gallbladder neck that could not be discriminated whether it was bile sludge or a polyp were revealed by ultrasonography. Computed tomography revealed a 32 × 33 mm mass lesion with soft tissue density and heterogenic contrast enhancement on the gallbladder neck (Figure 1) and horseshoe kidney anomaly (Figure 2). The blood test showed hypothyroidism due to Hashimoto's thyroiditis and increased carcinoembryonic antigen (CEA) 19.2 ng/ml and carbohydrate antigen-125 (CA-125) 62.2 IU/ml levels with normal carbohydrate antigen 19-9 (CA 19-9) < 0.8 IU/ml. Thyroid hormone replacement therapy was initiated after internal medicine consultation. After establishing euthyroidism the patient was hospitalized. We performed laparoscopic exploration and encountered a solid mass in the Calot region, so exploration was converted to laparotomy. The gallbladder was hydropic and the wall was intensely thickened. The solid mass that was revealed in laparoscopic exploration was possibly an infiltrated cystic lymph node. Cholecystectomy was performed and the gallbladder sent to the pathology department for frozen examination. The result was malign and the cystic resection margin was involved by the tumor. So we dissected the remnant cystic duct towards the choledococystic confluence and resected the remnant at the junction and sent the resected duct and paraaortocaval region lymph node sample for frozen section study to identify the extent of the disease. There was no

Corresponding author:

Durmuş Ali Çetin
Gastroenterological
Surgery Department
Kartal Koşuyolu
High Speciality and
Training Hospital
34650 Istanbul, Turkey
Phone: +90 5054378667
E-mail: drdurmusalicetin@

gmail.com



Figure 1. Mass lesion with soft tissue density and heterogenic contrast enhancement on gallbladder neck (marked with red arrow)

involvement in the lymph node samples, and the cystic duct remnant resection margin was free of tumor. According to these results further extensive radical surgery was performed and he underwent resection of the liver bed, hepatoduodenal lymph node dissection and port-site resection in the same operation session. Bile fistula developed in the early postoperative course but fortunately it was closed spontaneously by a conservative approach. He was discharged from hospital on the 12th day of surgery. As the patient was informed about the properties of his illness he agreed with us to report this condition and we obtained written consent from him. Resection margins were free of tumor and the disease was classified as stage IIIB (T3N1M0) according to the American Joint Committee on Cancer (AJCC) TNM Staging for Gallbladder Cancer (7th ed., 2010) system.

Gallbladder cancer is the fifth most common malignant neoplasm of the gastrointestinal tract and is associated with a poor prognosis. Gallbladder cancer is two-six times more frequent in women and is characterized by geographical and ethnic diversity [4].

Hundal and Shaffer declared in their article that from 2010 US data, age-adjusted incidence rates (per 100,000) in 2010 had risen from 0.16/100,000 (for those 20–49 years old) to 1.47/100,000 (for those 50–64 years old), to 4.91/100,000 (65–74 years), and to 8.69/100,000 for individuals over the age of 75 years [5].

There were no patients with gallbladder cancer, horseshoe kidney and Hashimoto's thyroiditis in the literature. Horseshoe kidney and Hashimoto's thyroiditis can be seen together in Turner syndrome occurring in women who are missing one X chromosome [6]. As our patient is male we cannot argue that the case is Turner syndrome. On the other hand, these anomalies could exist together with other unknown syndromes. Hashimoto's thy-



Figure 2. Computed tomography slice showing horseshoe kidney anomaly

roiditis is an autoimmune disorder [7]. Castro *et al.* concluded that a large number of autoimmune conditions were associated with increased risk of hepatobiliary tract cancer [8]. In their report, among 402,462 patients with autoimmune disorders, 330 were diagnosed with gallbladder cancer and 10 of them had Hashimoto's thyroiditis.

As a limitation, we do not have the genetic analysis of the patient and in our literature review we did not encounter such a triad that our patient harbors. So for now it remains a speculative query that is waiting for an answer, perhaps with more case series and biogenetic studies in the future.

Conflict of interest

The authors declare no conflict of interest.

References

- 1. Rakic M, Patrlj L, Kopljar M, et al. Gallbladder cancer. Hepatobiliary Surg Nutr 2014; 3: 221-6.
- Gündeş E, Tekin A, Ferlibaş E, Bal A, Esen HH. Gallbladder malignant lymphoma: a rare case. Dicle Med J 2013; 40: 492-94.
- 3. Duffy A, Capanu M, Abou-Alfa GK, et al. Gallbladder cancer (GBC): 10-year experience at Memorial Sloan-Kettering Cancer Centre (MSKCC). J Surg Oncol 2008; 98: 485-9
- Konstantinidis IT, Deshpande V, Genevay M, et al. Trends in presentation and survival for gallbladder cancer during a period of more than 4 decades: a single-institution experience. Arch Surg 2009; 144: 441-7.
- 5. Hundal R, Shaffer EA. Gallbladder cancer: epidemiology and outcome. Clin Epidemiol 2014; 6: 99-109.
- Bondy CA. Turner's syndrome and X chromosome-based differences in disease susceptibility. Gender Med 2006; 3: 18-30
- Khan FA, Al-Jameil N, Khan MF, Al-Rashid M, Tabassum H. Thyroid dysfunction: an autoimmune aspect. Int J Clin Exp Med 2015; 8: 6677-81.
- 8. Castro FA, Liu X, Forsti A, et al. Increased risk of hepatobiliary cancers after hospitalization for autoimmune disease. Clin Gastroenterol Hepatol 2014; 12: 1038-45.

e140 Arch Med Sci Civil Dis 2016