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Letter to the Editor

Krukenberg Tumor Manifesting with Hirsutism and Acanthosis Nigricans as the Exclusive Presenting Symptoms

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Dear Editor-in-Chief

Krukenberg tumor (KT) is a rare metastatic ovarian tumor defined as a carcinoma that consists of mucin-filled signet-ring cells in >10% of the tumor (1). Nevertheless, this histopathologic definition is not followed by all authors and the term KT is occasionally used inaccurately for all metastatic tumors to the ovary (1). Krukenberg tumor patients are usually young women with a mean age of 45 years (2). The most common origin of KT is the stomach (up to 70% of cases), followed by the colon and rectum (10%), the breast (5%), the gall bladder, and biliary tract (2%). The first manifestations of KT are usually abdominal pain and distension (1, 2). Abnormal uterine bleeding or alterations in menstrual period may be detected infrequently. There are few reports of pregnant women suffering from Krukenberg tumor in association with virilization or hirsutism in the literature (3-5).

Here, we report a 23-year-old woman with Krukenberg tumor manifesting by hirsutism and acanthosis nigricans without pregnancy from Tehran, Iran. The case is of special interest because it affected a non-pregnant woman and hirsutism and acanthosis nigricans were the initial manifesting symptoms which occurred prior to the diagnosis of Krukenberg tumor.

A 23-yr-old virgin woman presented to our Dermatology Clinic with the complaints of hirsutism on her chin, chest and limbs in addition to darkening and thickening of the periorbital and perioral skin, neck, groin, and axillae for about 8 months. She had no history of weight gain/ loss, disregulation of her menstrual period, and gastrointestinal symptoms. On physical examination velvety, hyperpigmanted plaques were noted around her lips and eyes, back of neck, inframammary folds, groin and axillae, compatible with acanthosis nigricans (Fig. 1). She also had hirsutism on her face, neck, intermammary cleft, and limbs (Fig. 2, 3). Histopathologic examination revealed hyperkeratosis and mild acanthosis undulating with dermal papillomathosis, thereby confirming the diagnosis of acanthosis nigricans (Fig. 4). Routine laboratory tests including hormonal evaluation were normal. Ultrasonography of abdomen and pelvis revealed bilateral ovarian solid masses in association with ascites and periaortic and mesenteric lymphadenopathy. Then laparoscopic biopsies of lymph nodes, mesenter and ovaries were performed which showed metastatic carcinoma with abundant signet-ring cells. Endoscopy along with histopathologic evaluation revealed the primary source of tumor which was a gastric adenocarcinoma (Fig. 5).

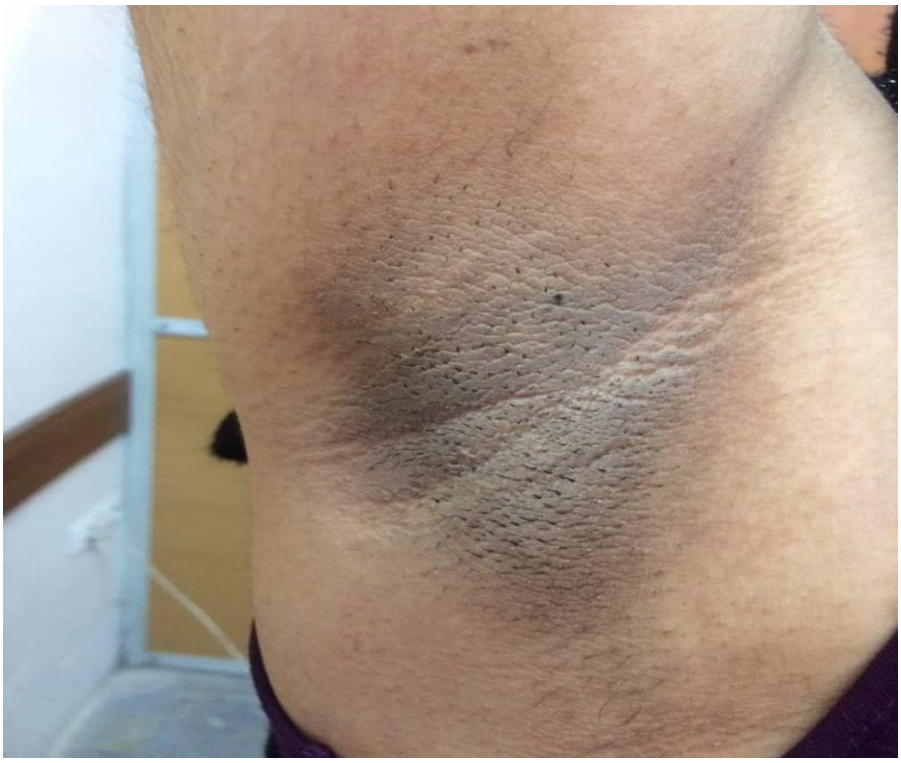

Fig.1: Acanthosis nigricans of axillae region of the patient

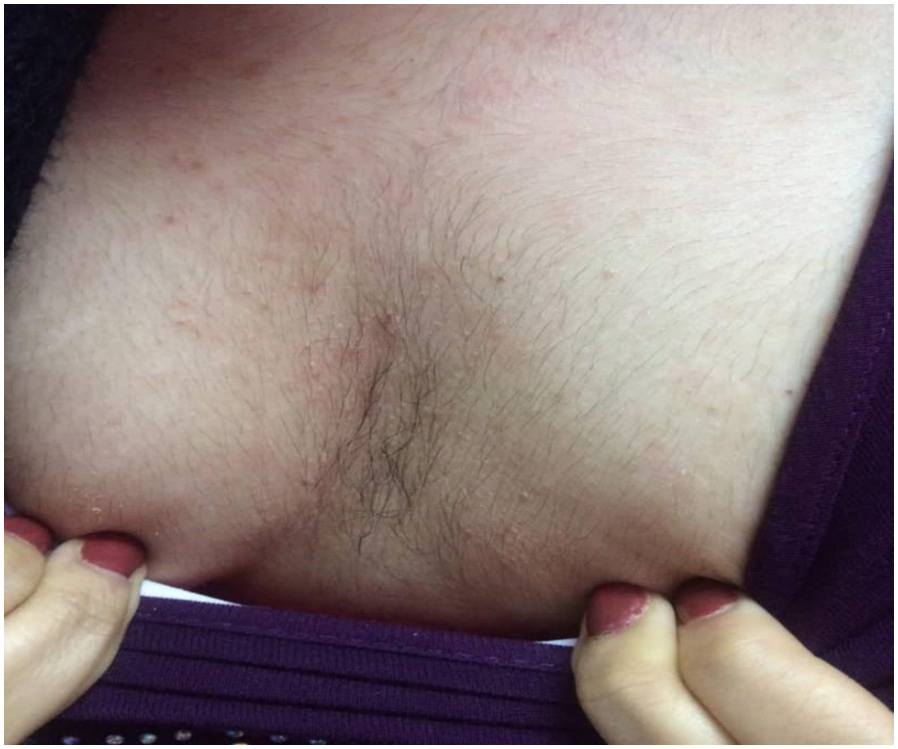

Fig. 2: Hirsutism of the intermammary cleft

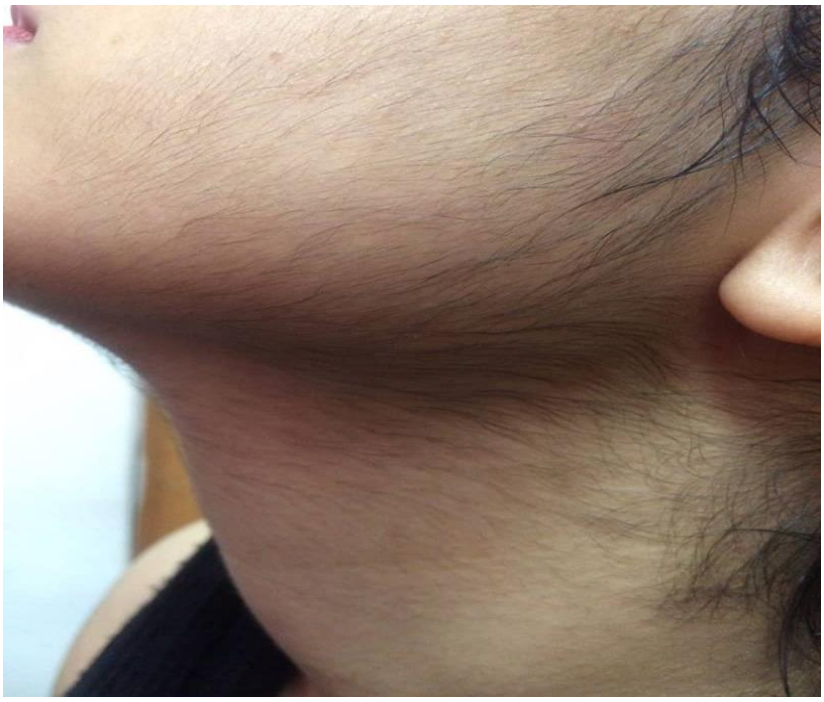

Fig. 3: Face, and the neck

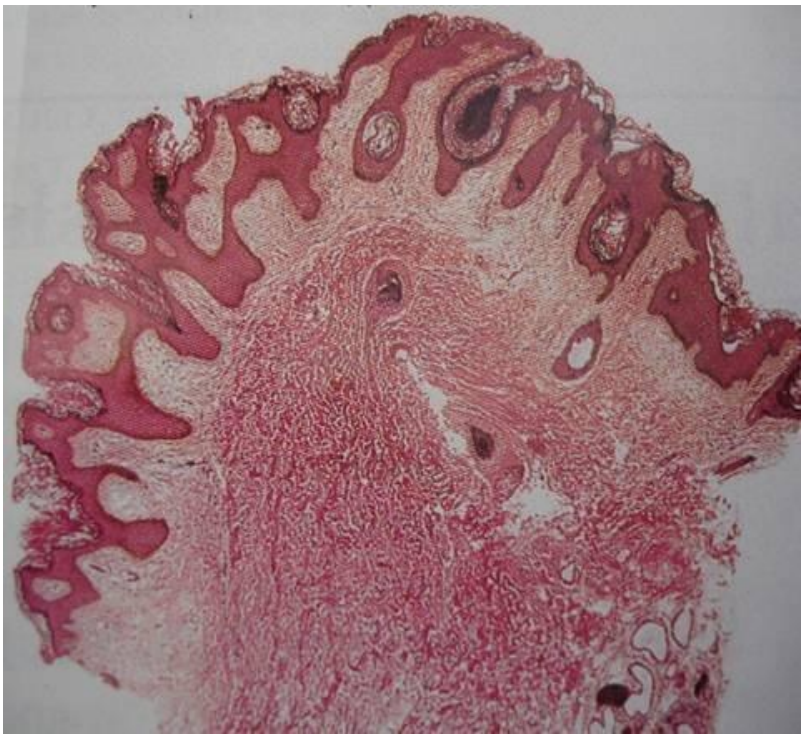

Fig. 4: Histopathologic examination of axillary lesion: hyperkeratosis and mild acanthosis undulating with dermal papillomathosis compatible with acanthosis nigricans (H & E×10)

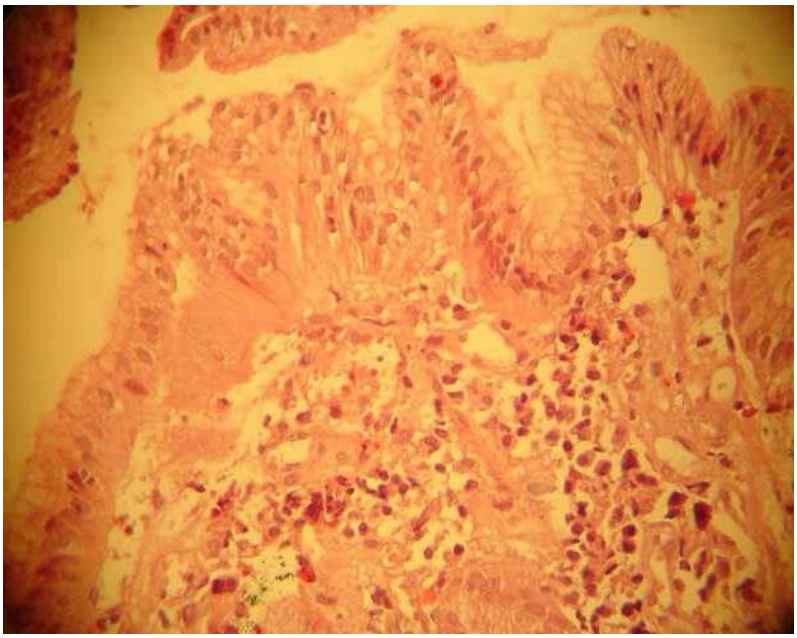

Fig. 5: Histopathologic examination of gatric tumor: gastric adenocarcinoma with signet-ring cells (H & E, ×400)

Surgery and chemotherapy were recommended for the patient, but unfortunately, the patient was lost for follow-up. Informed consent was taken from the patient before the report. Almost 6% of malignant adnexal tumors are metastatic (3). Krukenberg tumors are ovarian metastatic carcinoma that consists of mucin-filled signet-ring cells in >10% of the tumor (1). The most frequent presenting symptoms are abdominal pain and distension (2). Other probable symptoms of KT are virilization, hirsutism, breast tenderness and swelling, amenorrehea, and abnormal uterine bleeding which occurs only in 20% of cases (1, 2). In most cases, KT is diagnosed before the primary tumor and the detection of its origin remains challenging in many cases (1, 2). Two-thirds of primary tumors are discovered in the stomach. Other origins of KT could be colon and rectum, breast, gall bladder and biliary tract, small intestine, appendix, pancreas, uterus, bladder and renal pelvis (2).

In our case, the patient was 23 yr old, tumor was bilateral and primary tumor, which was a gastric adenocarcinoma, was diagnosed after ovarian metastasis. Although abdominal pain and distension are the most common manifestations of KT, in our case the only presenting manifestations were hirsutism and acanthosis nigricans. Rarely and surprisingly, abdominal pain/mass, nonspecific gastrointestinal signs, and fatigue were absent in our patient, as early presentations. The literature review conceals few reports of KT in association with hirsutism/virilization , but all of these cases were pregnant women. (3, 4). To our knowledge, there is only one report of virilization occurred in a non-pregnant woman due to KT in the literature; however, in that case virilization occurred 3 months after the diagnosis of KT (5). Therefore, our case is the first and the sole case of KT presenting with hirsutism in association with acanthosis nigricans as initial symptoms in a non-pregnant woman. The main treatment of KT is surgery. Prognosis of KT is very poor with a median survival of 7-14 months (2).

In spite of the rarity of the Krukenberg tumor, physicians should keep it in mind as a differential diagnosis in young women with hirsutism, acanthosis nigricans, and enlarged ovaries, since its misdiagnosis or late diagnosis would impose a

huge burden both to the patient and the health system.

Conflict of interest

The authors declare that there is no conflict of interests.

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