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Case Report

Heterotopic respiratory mucosa of the uterine corpus

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ABSTRACT

A 47-year-old, single, Chinese woman presented with pain on the 1st day of menses for more than 30 years. Her dysmenorrhea worsened over years and underwent a total abdominal hysterectomy and bilateral salpingo-oophorectomy. The myometrium showed trabeculated appearance, and there were adhesions between ovaries and fallopian tubes. A pale solid brownish mass measuring 1.5 cm × 1 cm × 0.6 cm with fibrous whitish cut surfaces was present on the lateral wall of the uterus in the lower uterine segment. Histologically, adenomyosis and left ovarian endometriosis were confirmed. The lateral uterine wall nodule showed a tubular structure lined by ciliated pseudostratified columnar epithelium. Smooth muscle bundles were found around the entire tubular structure. Lobules of salivary type glands containing both serous and mucous cells are present. The pathological diagnosis of heterotopic respiratory mucosa (HRM) was made. To our knowledge, this is the first reported case of HRM of the uterine corpus.

KEY WORDS: Heterotopia, respiratory mucosa, uterus

INTRODUCTION

Heterotopia refers to the presence of normal tissue in abnormal locations.Heterotopic tissues of the uterine corpus are usually an incidental finding in women of reproductive age group. The tissues are most commonly cartilage, bone, glia, and fat and are usually within the endometrium (including endometrial polyps), less commonly the cervix or myometrium. There are reports on heterotopic respiratory mucosa (HRM) in the rectum [1,2]. However; HRM of the uterine corpus has not been reported in the literature. We describe the first case of HRM of the uterine corpus in this report.

CASE REPORT

A 47-year-old, single, Chinese woman presented with pain on the 1st day of menses for more than 30 years. She had a history of heavy menses for more than 10 years. She underwent laparotomy for bilateral endometriotic ovarian cysts removal which was confirmed on histopathological examination. Her dysmenorrhea worsened over the years but was relieved by analgesics. Ultrasound examination revealed an echogenic mass on the posterior uterine wall measuring 5.4 cm × 5.4 cm, noted to be increasing in size on 2 yearly examinations. Subsequently, the patient underwent a total abdominal hysterectomy and bilateral salpingo-oophorectomy. She had an uneventful post-operative recovery and remains well following surgery.

On gross examination, an enlarged uterus with the cervix (11 cm × 8 cm × 5.6 cm) and bilateral tubes and ovaries weighing 340 g was received in 10% buffered formalin. Fibrous adhesions between the ovaries and fallopian tubes were found. A pale solid brownish mass (1.5 cm × 1 cm × 0.6 cm) with fibrous whitish cut surfaces was present on the lateral wall of the uterus in the lower uterine segment. The myometrium showed trabeculated appearance. Histological findings revealed adenomyosis of myometrium and endometriosis of left ovary. The lateral uterine wall mass showed a tubal structure lined by ciliated pseudostratified columnar epithelium [Figure 1]. Smooth muscle bundles were found around the entire tubular structure. Lobules of salivary type glands containing both serous and mucous cells were present [Figure 2]. Immunohistochemical studies for thyroid transcription factor-1 (TTF-1) and estrogen receptor (ER) were negative [Figure 3a and b]. Eventually, this structure was diagnosed as HRM.

DISCUSSION

Heterotopic tissues in the uterus are not always pregnancy related and are discovered usually in women of reproductive

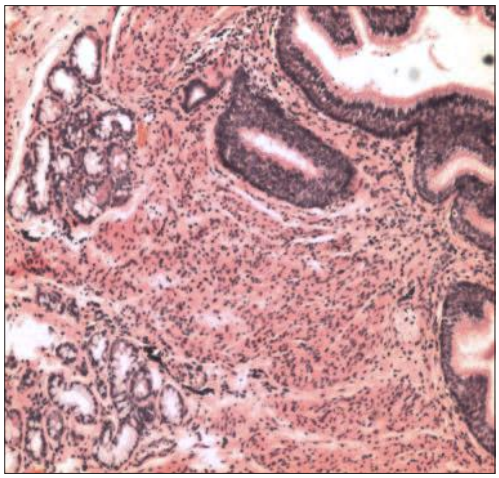

Figure 1: Photomicrograph showing a tubal structure lined by ciliated columnar epithelium (H and E, ×40)

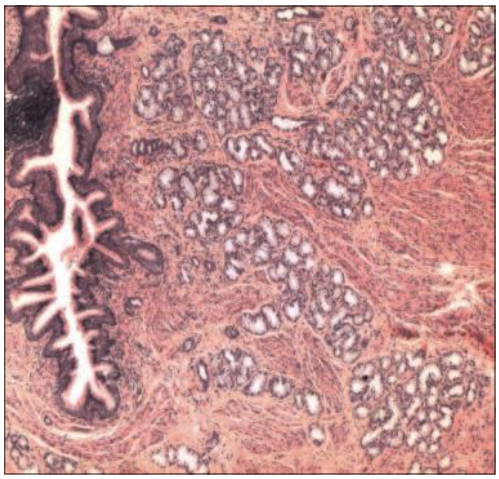

Figure 2: Photomicrograph showing lobules of seromucinous glands and smooth muscle bundles (H and E, ×20)

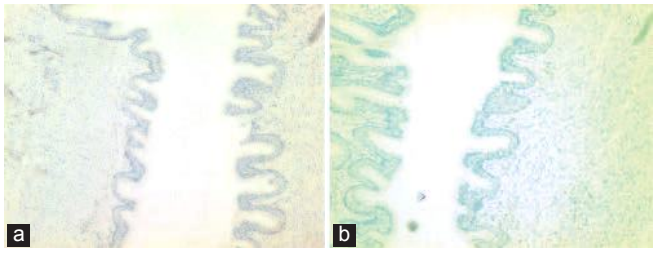

Figure 3: (a) Negative immunostaining for thyroid transcription factor-1 (×40). (b) Negative immunostaining for estrogen receptor (×40)

age group. Heterotopic tissues of glia, cartilage, and bone in the uterus have been reported in the literature. Glial heterotopia has been found in cervix and endometrium [3-5]. Glial tissue along with foci of bone, cartilage and squamous epithelium has been reported in both cervix and endomyometrium [6]. Glial heterotopia is usually an incidental finding during histopathological examinations on tissues submitted for another diagnosis [7]. Implantation of fetal tissue during therapeutic or spontaneous miscarriages is the presumed (and in some cases proven) pathogenesis [7,8]. Some tissues may be a result of true heterotopia, metaplastic process or due to dystrophic origin [8].

Heterotopic cartilage can be found in the uterus. The proposed mechanisms were metaplasia of multipotent stem cells the following trauma due to childbirth, hyperestrinism, hypercalcemia, and iatrogenic implantation of fetal tissues including cartilage into the uterine wall following dilatation and curettage [9-11]. Cases of endometrial osseous metaplasia have been reported. Heterotopia, dystrophic calcifications, and ossification of post-abortive endometritis, metastatic calcification, metaplasia in healing tissue, prolonged estrogenic therapy after abortion, and retained fetal bone are the commonly proposed mechanisms [12]. Our patient had no history of pregnancy or abortion.

HRM is a rare entity. There are two case reports on HRM in the rectum by Kunimitsu *et al*. and Mitsuaki Ishida *et al*. in 2007 and 2014, respectively [1,2]. Immunohistochemical studies for TTF-1 and ER were negative in the case reported by Mitsuaki Ishida *et al*. similar to the findings in our case [2]. Foregut heterotopia was the proposed concept by Kunimitsu *et al*. [1]. However, none of such cases have been reported in the uterus. Embryologically, the uterus is derived from mesoderm, and respiratory mucosa originates from endoderm. The mucosal lining of the respiratory tract forms as an out-pocketing from foregut (pharyngeal endoderm) and glands arise as endodermal out pocketing at various points. The histogenesis of respiratory mucosa on the uterine corpus remains unclear from the embryological point of view. Hence, the possibility of ectopic respiratory mucosa as a result of foregut heterotopia has to be considered in this case similar to that of the rectum.

CONCLUSION

To our knowledge, this is the first case of HRM to be reported in the uterus. Pathologists should be aware of this rare entity and can easily diagnose when the possibility of HRM is thought of in relevant circumstances.

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