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Case Report

Uterine cervical mesonephric hyperplasia with focal cystic change masquerading clinicopathologically as lobular endocervical glandular hyperplasia to malignancy

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ABSTRACT

Introduction: Although uterine cervical mesonephric hyperplasia (MH) arising from mesonephric remnants is a well-known but uncommon entity, it might pose a clinicopathological diagnostic challenge to distinguish from lobular endocervical glandular hyperplasia (LEGH) or adenocarcinoma, if MH rarely presents as a lobular and/or cystic mass with expansion of the cervical wall. However, few papers have described the detailed clinicopathological characteristics of MH compared to those benign to malignant lesions.

Case presentation: An early forties Japanese female presented with a history of increased vaginal watery discharge. Multiple cystic lesions measuring less than 3 mm in diameter generated a high signal intensity on T2-weighted MRI, in the bilateral aspects of the variably enlarged uterine cervix. A gross examination of a hysterectomy specimen revealed bilateral small multicystic lesions, filled partly with secreted fluids, measuring approximately 25 × 7 × 5 mm, respectively, located in the superficial to relatively deep cervical wall. A microscopic examination showed that these lesions predominantly consisted of a lobular proliferation of small to medium-sized tubules and cysts containing occasionally periodic acid-Schiff-positive eosinophilic/pink secreted materials, lined by bland-looking cuboidal to flattened epithelium. Immunohistochemically, these lining cells were specifically positive for CD10 in a characteristic luminal staining pattern, whereas negative for MUC6, and had a low MIB-1 labeling index. We ultimately made a diagnosis of cervical MH, lobular type, with focal cystic change.

Conclusion: We should be aware that, since gynecologists/pathologists might misinterpret MH as LEGH to malignancy, including the malignant counterpart of MH, a wide panel of immunohistochemical antibodies can be helpful supplemental tools.

1. Introduction

Particularly in women, the mesonephric (Wolffian) ducts embryologically regress during their development, however, if persistent, those remnants might be recognized incidentally even in the lateral walls of the uterine cervix [1–4]. Mesonephric remnants were first described by Meyer in 1907 [3,4], and subsequently, Ferry and Scully reviewed their experience, in which mesonephric remnants comprised up to 20% of all the cervical specimens largely depending on the methods of sampling [3,5]. Hyperplasia of these elements, i.e., uterine cervical mesonephric hyperplasia (MH), is a well-known established entity but occurs uncommonly, and MH not only involves the deep layer of the cervical wall, but also extends close to the luminal surface intermingled with pre-existing endocervical glands [3,4]. Although generally asymptomatic, if MH rarely presents as a lobular and/or cystic mass with expansion of the cervical wall, it might pose a clinicopathological diagnostic challenge to distinguish widely ranging from lobular endocervical glandular hyperplasia (LEGH) to malignancy, such as mesonephric carcinoma or minimal deviation adenocarcinoma (MDA) [6,7]. However, few papers have described the detailed

Abbreviations: MH, mesonephric hyperplasia

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clinicopathological features of MH compared to those benign to malignant lesions.

Histopathologically, MH of the uterine cervix has been sometimes classified into several types, based on the architectural pattern of the glands, even though there would be no clinical significance [3,4]. The lobular MH is the most common type, characterized by a lobular arrangement of clustered, small to medium-sized and round, but occasionally dilated cystically, mesonephric tubules filled with periodic acid-Schiff (PAS)-positive eosinophilic to pink amorphous materials, often arising deeper in the cervical stroma and separated by variable amounts of stroma [3]. The less common type is called the diffuse MH, characterized predominantly by a non-clustered, extensive and diffuse proliferation of mesonephric tubules separated by varying amounts of cervical stroma, coexisted occasionally with minor foci of the lobular MH [3]. Finally, the least common type of MH is the duct MH, composed of one or more ducts lined by hyperplastic-appearing epithelium without atypia, usually lacking the intraluminal eosinophilic/pink secretions and often displaying clefted contours [3].

We herein report a rare case of lobular type MH with focal cystic change originating from the bilateral aspects of the variably enlarged uterine cervix, showing unique clinicopathological features and masquerading as wide range from LEGH to malignancy. The first clinicopathological examinations did not allow for a conclusive diagnosis, and the final diagnosis was ultimately made based on a wide panel of immunohistochemical analyses, including its hallmark antibodies, CD10, MUC6 and MIB-1.

2. Case report

An early forties Japanese female presented with a history of increased vaginal watery discharge. She had an unremarkable medical history, except for ventricular septal defect in childhood. The laboratory data, including the blood cell count, chemistry, and tumor markers, were within normal limits. Localized multiple cystic lesions measuring less than 3 mm in diameter generated a high (white to white-grayish) signal intensity on T2-weighted MRI images of both sagittal and horizontal sections, in the bilateral aspects of the mildly enlarged uterine cervix (Fig. 1A). A routine cervical smear detected no apparent atypical cell clusters. The gynecologists first interpreted these lesions as benign, such as a LEGH, but could not completely rule out malignancy.

Simple total hysterectomy was performed, and a gross examination of its cut surface revealed bilateral small multicystic lesions, filled partly with secreted clear fluids, measuring 25 × 7 × 5 mm (lt. side) or 23 × 7 × 4 mm (rt. side), respectively, located in the superficial to relatively deep cervical wall variably thickened (Fig. 1B). On scanning magnification, these cervical lesions showed a well-circumscribed and localized multicystic-like foci, measuring less than 3 mm in diameter of each cyst or tubule (Fig. 1C). Resection was diagnosed as complete by this histopathological examination.

Microscopically, these lesions predominantly comprised a lobular proliferation of small to medium-sized, non-complex tubules with focal cystic change (Fig. 2A), containing occasionally PAS-positive eosinophilic/pink, amorphous and secreted materials, lined by non-ciliated, one-layered cuboidal to flattened epithelium having bland nuclei and no cytoplasmic mucicarmine-positive mucin without any evidence of mitotic figures (Fig. 2B). They extended to not only middle deep layer of the cervix, separated by variable amounts of stroma, but also close to the cervical mucosal surface with focal cystic change (Fig. 2A & B). There were neither desmoplastic reaction to invasive growth nor periglandular stromal edema. Furthermore, pre-existing endocervical glands and endometrium/myometrium have no remarkable change. In immunohistochemistry, the lining cells of tubules/cysts were specifically positive for CD10 in a characteristic luminal staining pattern (Fig. 2C), whereas completely negative for MUC6 and estrogen receptor (ER), and demonstrated a low Ki67 (MIB-1) labeling index (1 to 3%) (Fig. 2D). In a background, immunohistochemically p16-positive cells were absent.

Based on these features, we finally made a conclusive diagnosis of MH, lobular type, with focal cystic change, arising from the bilateral aspects of uterine cervix. To date, after approximately 1 and half years of post-operative follow-up, the patient remains well without any evidence of recurrence/metastases.

3. Discussion

More recently, very few case reports and review articles available on uterine cervical MH have suggested that a confident and accurate diagnosis might be impossible based on a clinical and morphological (H&

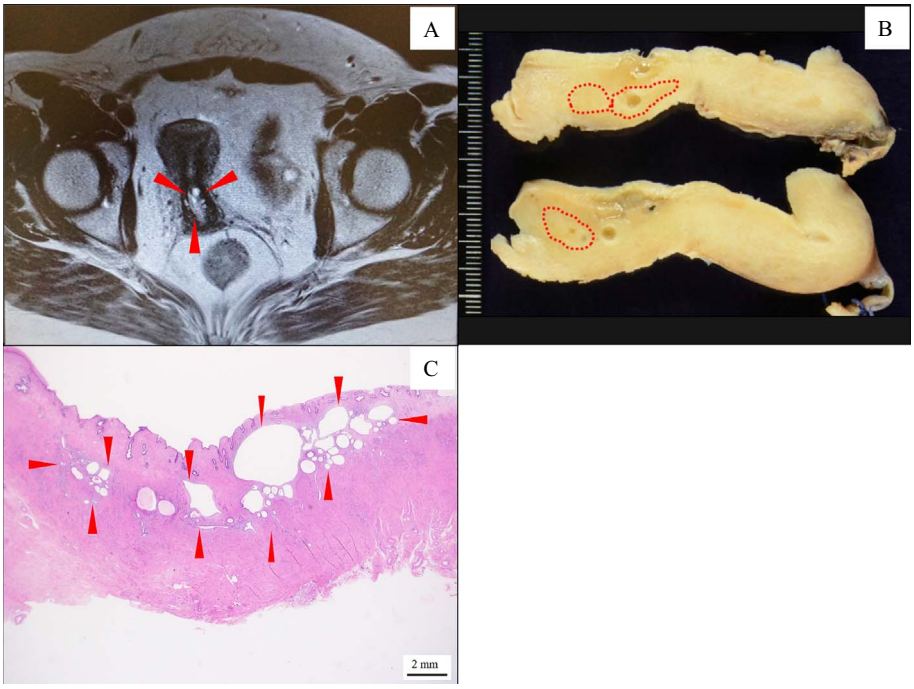

Fig. 1. The imaging, gross and loupe findings at surgery of the uterine cervical MH. (A) Localized multiple cystic lesions measuring less than 3 mm in diameter generated a high (white to white-grayish) signal intensity (arrowheads) on T2-weighted MRI images of horizontal sections, in the bilateral aspects of the mildly enlarged uterine cervix wall. (B) On the cut surface from the hysterectomy specimen, gross examination showed bilateral small multicystic lesions, filled partly with secreted clear fluids, measuring approximately 25 × 7 mm, respectively (dot areas), located in the superficial to relatively deep cervical wall variably thickened. (C) On scanning magnification (H&E stain), these cervical lesions demonstrated a well-circumscribed and localized multicystic-like foci (arrowheads), measuring less than 3 mm in diameter of each cyst or tubule. Bar = 2 mm.