

POLYPOID TERATOMA OF THE UTERINE CERVIX

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Abstract

Reported is a case of 16 year-old girl with a huge polypoid tumor of the uterine cervix comprising 10 or more nodules and filling the vaginal lumen. The gross appearance of the tumor resembled that of a botryoid sarcoma.

Histological study indicated that the tumor consisted largely of myxoid mesenchymal tissues, including foci of undifferentiated mesenchymal cells, cartilage and striated muscles with various degrees of maturation, smooth muscles, possible respiratory epithelium and neuroglial tissues in addition to many microcystic structures like Nabothian follicles.

She has been free of recurrence postoperatively for 5.5 years. The tumor is interpreted on these data as representing a benign teratoma of the uterus, which is of extreme rarity and probably the first reported case in Japan.

It is emphasized that the uterine teratoma must be distinguished from botryoid sarcoma and benign papilloma or polypoid tumor of the uterine cervix because of the similarity of the gross appearance.

Teratoma of the uterus is an extremely rare tumor. Only 6 cases have been accepted since the original report made by Mann in 1929,¹⁾⁻⁶⁾ and none of the case has been reported in Japan as far as we know.

In this paper a case of uterine teratoma with polypoid appearance is reported and the differential diagnosis for the disease is assessed because of a possible confusion with botryoid sarcoma and others.

CASE REPORT

A 16 year-old girl was first seen in May of 1969, with an approximate 1 month's history of bloody vaginal discharge. She had had an appendectomy about 1 month ago because of a headache, pyrexia, abdominal pain and the genital bleeding, but the bleeding persisted postoperatively and again she had suffered an abdominal pain and pyrexia about a week ago. The menarche was at the age of 11 years and the men-

Foot-note: A part of this study was presented at The 59th Annual Meeting of The Japanese Pathological Society, 1970, Kyoto.

strual periods were irregular. The past history was not eventful except for the appendectomy.

On physical examination the patient was well-nourished, but the palpebral conjunctiva appeared considerably anemic.

On vaginal examination a huge tumor mass with multi-polypoid appearance was found filling up the vaginal lumen and portions of it were exposed to the external genitalia, showing a focal hemorrhage and granulation (fig. 1).

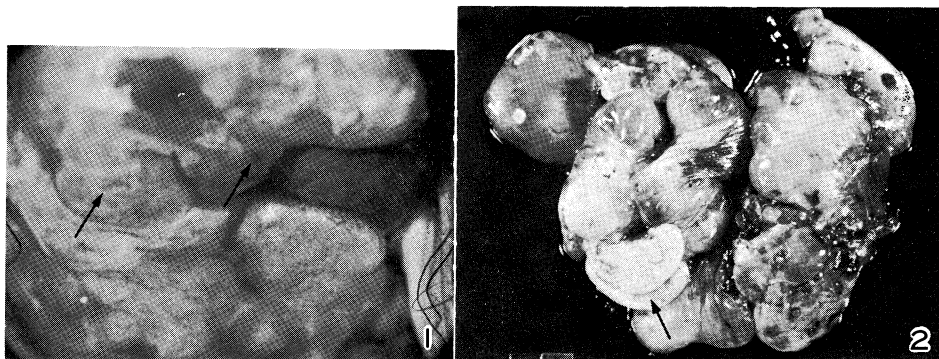


Fig. 1. The multipolypoid or botryoid tumor mass being exposed to the external genitalia and showing a focal hemorrhage and granulation (arrows).

Fig. 2. The resected tumor mass comprising 10 or more and mostly jelly-soft nodules connected each other. A few nodules are hard and fibrous resembling the ovary (arrow).

Laboratory data included: RBC 354×10^4 , WBC 5,500 with normal analysis, Hb 9.0 g/dl, Platelets 26×10^4 , Ht 32 %, Serum syphilis reaction(-), Serum total protein 8.0 g/dl with normal fractionation, NPN 33 mg/dl, BUN 15 mg/dl, Creatinine 0.5 mg/dl, Total cholesterol 210 mg/dl, Cholesterol ester 74 mg/dl, Choline esterase pH=0.61, Takata reaction(-), Gros reaction(-), CCLF(-), Serum cobalt reaction $R_{4(5)}$, Icterus index 4.0, GOT 21 u., GPT 14 u., Alkaline phosphatase 6.5 u. Normal urinalysis.

The biopsy disclosed only an inflammatory granulation with remarkable edema and thus it was not diagnostic.

At total excision of the tumor mass, the tumor measured approximately an adult fist-size in over-all dimension, comprising 10 or more thumb tip- to walnut-sized nodules connected each other like a bunch of grapes (fig. 2), and it was pedunculated with an approximately 3 mm-thick stalk from the posterior lip of the portio vaginalis. The nodules were generally soft like a jelly, but a few of them were hard and solid.

On section the majority of the nodules appeared microcystic like Nabothian follicles and the soft portions grayish white, edematous or myxoid while the hard portions appeared white and fibrous, resembling the appearance of ovaries (fig. 3).

HISTOLOGICAL FINDINGS

The soft nodules are covered by a flattened stratified squamous epithelium, some portions of which are deeply invaginated, resulting in a pseudocystic structure. They consist of edematous or myxoid mesenchymal tissues with embryonal appearance, in which a focal proliferation of undifferentiated mesenchymal cells is found scattering (fig. 4). Areas

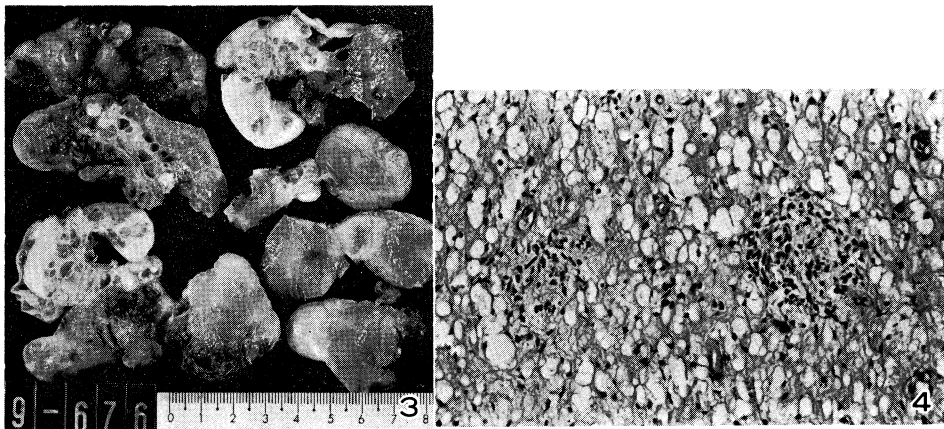


Fig. 3. Sections of the tumor nodules showing a myxoid and/or microcystic appearance.

Fig. 4. Myxoid mesenchymal tissues composing the soft tumor nodule and including a focal proliferation of undifferentiated mesenchymal cells. H & E, $\times 130$

of embryonal cartilage tissues, collagen fibers and striated muscles are also included in places and they show various degrees of maturation (fig. 5-8). These mesenchymal components, both mature and immature, by no means reveal any atypicality or pleomorphism. No tendency to focal aggregation of the undifferentiated mesenchymal cells as well as others is present underneath the covering squamous epithelium. The microcysts, grossly identified, are lined by a single layer of a flattened glandular epithelium and their lumina are highly distended by the retention of mucinous secretions. They are surrounded by fibrous stromas and indistinguishable from Nabothian follicles occurring frequently in the exocervix. Focally scattered are glands with multiple

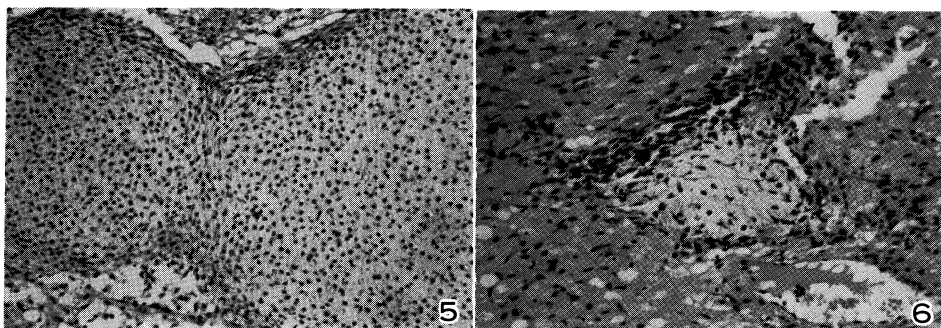


Fig. 5. Embryonal cartilage in the background of the mesenchymal tissue of the soft tumor nodule. H & E, $\times 130$

Fig. 6. Immature collagen fibers being formed within a focus of the undifferentiated mesenchymal cells. H & E, $\times 130$

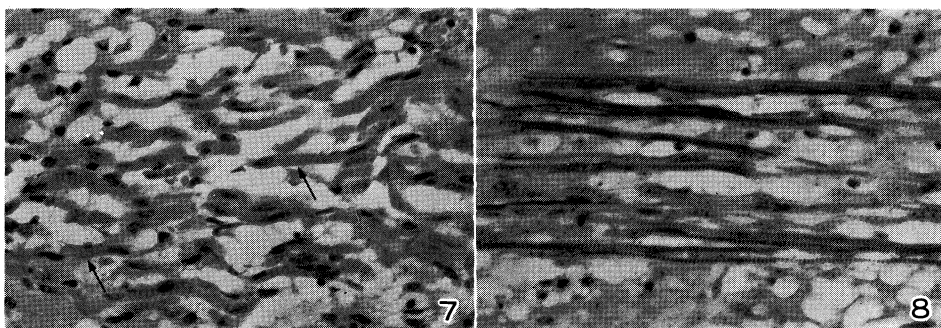


Fig. 7. A focus of immature striated muscles showing an occasional and indistinct striation (arrows). H & E, $\times 130$

Fig. 8. A focus of mature striated muscles showing definite striations. H & E, $\times 130$

branching and lining of a high columnar and clear epithelium. They appear similar to the normal cervical glands. A few immature glandular structures are found to be surrounded by irregular bundles of smooth muscle fibers. Occasionally the covering squamous epithelium is transformed abruptly into a glandular epithelium, producing a definite gland with multiple branching (fig. 9). These glands are lined by a high columnar ciliated epithelium from the corpus portion downwards (fig. 10) and partly surrounded by a layer of the cartilage (fig. 9). They altogether are very suggestive of the differentiation to a respiratory epithelium. Very occasionally found are areas with distinct border and resembling neuroglial tissues (fig. 11). The cells of these areas actually stain well with Cajal's glial cell staining (fig. 12).

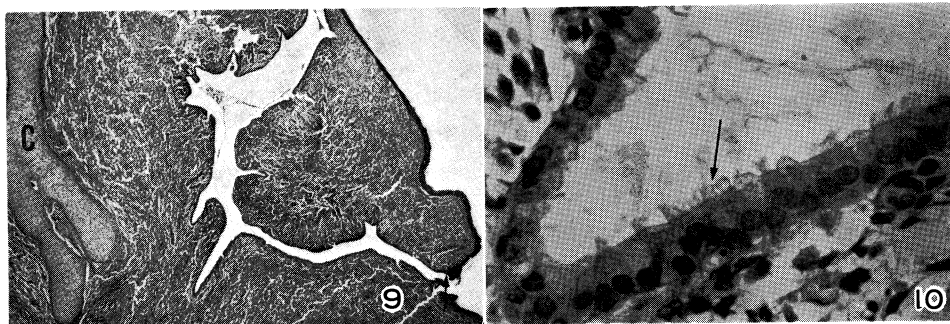


Fig. 9. The covering squamous epithelium transforming abruptly into a glandular epithelium (at the point of arrow), which forms a gland with multiple branching and is partly surrounded by a layer of cartilage (C). H & E, $\times 26$

Fig. 10. The glandular epithelium in Fig. 9 revealing definite cilia over wide areas (arrow). H & E, $\times 520$

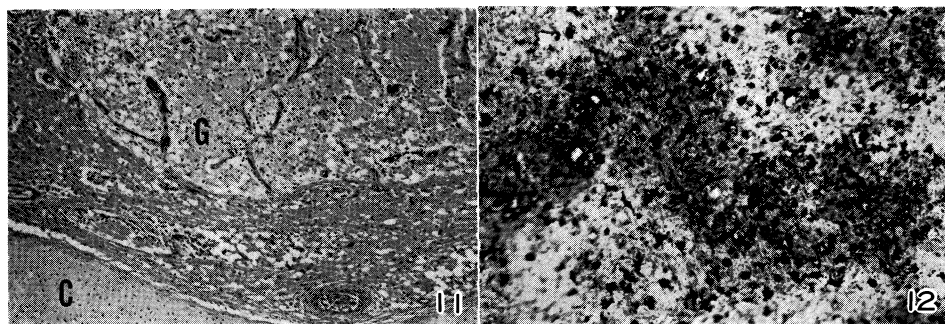
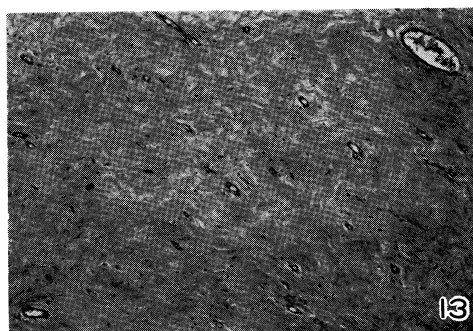


Fig. 11. A sharply bordered area simulating a neuroglial tissue (G). C: cartilage. H & E, $\times 52$

Fig. 12. The neuroglia-looking tissue including numerous cells stained well with Cajal's glial cell staining. Frozen sectioning, $\times 260$

The hard nodules are similarly covered by a stratified squamous epithelium and consist entirely of a fibrous mass comprising mature collagen fibers and of abundant small blood vessels (fig. 13).

Fig. 13. A vasculo-fibrous mass composing the hard tumor nodule. H & E, $\times 52$



CLINICAL COURSE

Although the tumor was considered histologically to be of benign nature because of the absence of sarcomatous change in the tissue components, the clinical findings including the gross appearance and location would have been suggestive of malignancy, particularly of a botryoid sarcoma and a simple hysterectomy was performed some days after the tumor excision. At operation only a bean-sized retention cyst was found located on the left ovary and otherwise nothing remarkable was present in the uterus, adnexae and in the pelvic cavity. Histological examination on the uterus disclosed no residual tumor involvement, and only a small submucosal abscess and vascular dilatation in the surrounding tissue were present in the posterior lip of the portio vaginalis from which the tumor was supposed to be pedunculated. The patient has been doing well postoperatively for five and half years, without any recurrence and metastasis at all.

DISCUSSION

Desite the botryoid appearance of the present tumor that may resemble the gross finding of botryoid sarcoma, the tumor components including the undifferentiated or embryonal mesenchymal cells lack the cellular stypism as sarcomas and an invasive nature. There was no tendency to focal cellular aggregation underneath the covering epithelium as often seen in botryoid sarcoma. Though single-layered, the ciliated columnar epithelium, in combination with the surrounding cartilage layer, appears considerably organoid and is highly suggestive of the differentiation to a respiratory epithelium. It seems rather evident that the tumor also contains neuroglial tissues. And probably most importantly the patient has been free of recurrence and metastasis for five and half years after the operation. Therefore, it seems reasonable to assume that the present tumor is of benign character and represents a teratoma comprising derivatives of all 3 germ layers.

Teratoma is defined by Willis⁷⁾ as a true neoplasm consisting of multiple kinds of tissues foreign to the part in which it arises. It can be mature or immature, cystic or solid, and benign or malignant. Derivatives of all 3 germ layers are usually included and at least 2 such derivatives should be identified before a teratoma can be confirmed. Tissue components of the 7 reported cases of uterine teratoma including the present case are listed in Table I, together with other informations. The patients appear to be at the age of reproduction, ranging from 16

TABLE I
Reported Cases of Uterine Teratoma

Author	Age	Parity	Site	Size & Shape	Description	Other findings
Mann (1922)	31	1/1	left tubal angle	apple-size	solid—skin, hair—follicles, sebaceous glands, tubular glands, smooth muscle, cartilage	
Hellendall (1930)	45	2/2	right tubal angle	hen-egg-size	cystic—skin, hair, sebaceous glands, cartilage, bone	left hydrosalpinx
Lackner and Krohn (1933)	34	2/2	cervical canal	4 cm, nodular pedunculated	solid—cartilage, smooth muscle, skin, sebaceous glands, hair, nerve cells and fibers, interstitial bronchial epithelium	
Forster (1952)	32	?	cervix vaginal vault	plum-size 2 nodules pedunculated hazel nut-size	microcystic—skin and appendages, smooth muscle, pyloric glands, pancreas	
Nicholson (1956)	38	0/0	uterine cavity	up to 6×4×3 cm 10 nodules 3.8 cm in dia. polypoid pedunculated	cystic—skin, hair, glandular elements, respiratory epithelium, hyaline cartilage, pancreas, bone, nerve	
Pyrah and Redman (1968)	33	3/3	cervical canal	5×3×2.5 cm 4.5×3×2.5 cm bilobulated	solid and microcystic—skin and appendages, cartilage, bone, nerve, smooth muscle, respiratory epithelium	2×1 cm pelvic teratoma partial uterus duplex
Present report	16	0/0	portio vaginalis	adult-fist-size 10 or more nodules with polypoid appear. pedunculated	edematous, solid, microcystic—undifferentiated mesenchymal cells, cartilage, striated muscle, smooth muscle, glandular epithelium, respiratory epithelium, neuroglial tissue	left ovarian retention cyst

TABLE II
Differential Diagnosis of Uterine Teratoma

	Age	Site	Gross appearance	Tissue component	Atypism	Prognosis
Teratoma	16 y	cervix	polypoid	1. myxoid tissue, cartilage, bone, striated & smooth muscles, etc.	none	
	45 y	corpus	botryoid lobulated pedunculated	2. tubular, respiratory, & digestive epithelium, pancreas, etc. 3. skin, sebaceous glands, hair, nervous tissues 4. organoid pattern(+) 5. focal cellular aggregation under covering (-)	none none	good
Botryoid sarcoma (Malignant Müllerian tumor, Malignant mesodermal mixed tumor)	newborn	urethra		1. myxoid tissue, cartilage, bone, striated & smooth muscle, fatty tissue, blood vessel, etc.	remarkable	
	juvenile	bladder	botryoid			
	adult	vagina	polypoid	2. glandular epithelium squamous epithelium		bad
	old age	cervix	pedunculated	3. organoid pattern(-) 4. focal cellular aggregation under covering(+)	remarkable at corpus	
Benign papilloma (Benign polypoid tumor 8), 9)	infant	cervix	papillomatous	1. paillary, fibrous or myxoid cores lined by columnar and/or squamous epithelium		
			polypoid pedunculated	2. no mesenchymal component 3. organoid pattern(-) 4. focal cellular aggregation under covering(-)	none	good

to 45 years of age in which our patient is the youngest. Sites of the tumor are at the cervix in 4 cases in which one is associated with a similar vaginal tumor, the tubal angle in 2 cases and the uterine cavity in 1 case. The tumors are usually large in over-all dimension and reported to measure from hazel nut- to apple-size. They are described as pedunculated in 4 out of the 7 cases and consisting of more than 2 nodules in 4 cases.

In differential diagnosis a botryoid sarcoma and benign papilloma or polypoid tumor of the uterine cervix may have to be distinguished from this disease, since these 3 lesions could equally disclose a polypoid or botryoid appearance. Findings for the differential diagnosis among the three may be summarized as in Table II.

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