

**Case Report**

# The Smallest Adenomatoid Tumour of Fallopian Tube Ever Reported

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**ABSTRACT**

A 26-year-old woman underwent bilateral tubectomy. Gross examination was unremarkable. On microscopic examination, a tumour was seen in one of the tubes, which measured 1 mm across. The tumour was located between the epithelial and the muscle layers and was composed of tubules lined by cuboidal to low columnar to flattened cells. The morphology was that of adenomatoid tumour. Subsequent sections did not show any tumour. This poses difficulty in evaluating the nature of the lesion, due to the inability to do stains for mucin and immunohistochemistry. The adenomatoid tumour reported in this article is the smallest of all those which have been reported in the literature. Further, this article discusses about the histogenesis of this tumour.

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**Introduction**

Adenomatoid tumour is the most common type of benign tumour of fallopian tube<sup>1</sup>. The name 'adenomatoid tumour' was given to this type of tumour as the tumour cells are arranged in a gland like pattern and resemble epithelial cells<sup>2</sup>. It is often asymptomatic and is detected incidentally during gross or microscopic examination of specimens removed for other lesions<sup>2-9</sup>. The other sites of occurrence of this tumour are uterus, ovary, paraovarian tissue, culde-sac,

testis, tunica vaginalis of testis, epididymis, adrenal gland<sup>1,2,5-7,9-21</sup>. The tumour tends to be small in size<sup>1-5,8,13,17</sup>, but can sometimes attain large size<sup>5</sup>. The histomorphologic, immunohistochemical and ultra-structural characteristics point towards mesothelial origin and is considered a type of benign mesothelioma confined to the genital tract<sup>1,5,6,10,11,14,17,22,23</sup>. We report a case of adenomatoid tumour which was not visible grossly, but made out only on a single



microscopic section, measuring only 1 mm across.

### Case report

A 26-year-old asymptomatic woman underwent puerperal sterilization at Chettinad Hospital and Research Institute, Tamilnadu, India. There was no history of abdominal trauma or pelvic inflammatory disease. Bilateral tubal segments were submitted in formalin for histopathological examination. Gross examination was unremarkable. Both the tubes were completely embedded. The sections were processed routinely and stained with Haematoxylin & Eosin (H&E). Microscopic examination revealed a circumscribed tumour in one of the tubes, measuring 1 mm across (**Fig. 1**), with ill-defined margins, between the epithelial and the muscular layers of the fallopian tube (**Fig. 2**). It was composed of tubules lined by cuboidal to low columnar flattened cells (**Fig. 3**). The lining cells were benign-looking. The lumina of the tubules did not show any secretion. The intercellular borders were indistinct forming a syncytium at many foci. Few lymphocytes and plasma cells were seen infiltrating the stroma surrounding the tubules, as an evidence of chronic inflammation. Special stains to demonstrate mucin and immunohistochemistry could not be done, as the tumour was not present in subsequent sections. There was no demonstrable continuity between the tumour cells and the serosal mesothelium or between the tumour cells and the tubal epithelium. The case was diagnosed as adenomatoid tumour.

### Discussion

Adenomatoid tumour of the female genital tract presents in patients between the ages of 26 and 55 years<sup>5</sup>. Our case belongs to this age group. It is more frequently

encountered in the uterine corpus than in the fallopian tube<sup>5,14,18</sup>. If present in the fallopian tube, the tumour presents like a nodule on examination of the external surface of the tube<sup>3</sup> ranging in size from 0.5 to 8 cm<sup>1-3,5,11,13</sup>. In our case, the tube was grossly unremarkable. The tumour was incidentally found on microscopic examination only.

There are many theories about the histogenesis of this tumour type.

The term adenomatoid tumour points to an epithelial origin, because, this term was coined by Golden and Ash, who felt that the cells of this tumour are epithelial<sup>2</sup>. Ragins and Crane<sup>3</sup> suggested the name of adenoma/ tubular adenoma. The presence of cytoplasmic vacuoles and the gland-like arrangement of the cells were considered to be in favour of epithelial origin. But Evans<sup>10</sup> feels that the morphology, location of the tumour, anatomical and histological relationships are not consistent with any of the recognized epithelial structures. Further epithelial specific markers such as Ber-EP4, B72.3, MOC-31, ER, PR are negative in this tumour<sup>1</sup>.

Theories of endothelial and mesonephric origin lost their popularity<sup>24</sup>. The cells lining the tubules are not spindle shaped. RBCs have never been found in the lumina of the tubules. Presence of abundant microvilli and few micropinocytic vesicles, dilated intercellular spaces<sup>11</sup>, absence of factor VIII related antigen, CD31 and CD34, positivity for keratin<sup>18,22</sup> are against the postulation of endothelial origin. Failure to explain the facts that no single adenomatoid tumour originated from the broad ligament or along the course of the Gartner's duct; tumour cells showed no resemblance to the epithelium lining the epididymis or the fallopian tube; failure to demonstrate any relationship between the tumour cells and the vestigial mesonephric structures in the

broad ligament or the epididymis preclude the theory of mesonephric origin<sup>24</sup>.

Theory of Müllerian origin was postulated by only a few authors. Honoré and O'Hara feel that location of the tumour within strictly müllerian territory is an indirect evidence for the müllerian origin and they further suggest the name of müllerian mesothelioma for this tumour<sup>25</sup>. Yasijima and Saito reported eight cases of adenomatoid tumours in the male genital tract. They describe two types of adenomatoid tumour differing in their histogenesis; out of which, the adenomatoid type is said to arise from the immature müllerian mesenchyme<sup>26</sup>.

The most consentient theory is of mesothelial origin<sup>1,5,6,10-12,14,17,20,21</sup>. Presence of direct continuity between the tumour cells and the serosal mesothelium, ultra-structural characteristics such as well-developed microvilli at the apical surface, abundant intracytoplasmic filaments, multiple desmosomes to which tonofilaments were attached, numerous cytoplasmic organelles<sup>5,10,23</sup>; histochemical characteristics such as presence of elevated concentrations of acid mucopolysaccharide in the lumina of the tubules and in the tumour cells<sup>1,11</sup>, immunohistochemical characteristics such as positivity for Cytokeratin, Calretinin, HBME-1, Vimentin, D2-40, WT-1, CK 5/6<sup>1,8,17</sup>; negativity for ER, PR, EMA<sup>6,7</sup> are in favour of mesothelial derivation of this tumour.

Jackson<sup>24</sup> suggests a theory of "Retrograde metaplasia". He explains that the Müllerian mesenchymal tissue undergoes metaplasia and differentiates into epithelial-like structures and hence he prefers to use the term "Benign müllerian mesenchymoma" to refer to this tumour.

Zubair A *et al*<sup>7</sup> states that the adenomatoid tumour arises from the pluripotent mesenchymal cells that

differentiate towards submesothelial cells and eventually mesothelial cells.

Ferenczy *et al*<sup>11</sup> and Vang and Wheeler<sup>1</sup> use the term "Benign mesothelioma" to refer to this tumour.

Davy and Tang suggest that the term "Adenomatoid tumour" should be restricted to light microscopic diagnosis; and the names "Adenomatoid mesothelioma" and "Adenomatoid angioma" be used when there is ultra-structural evidence for mesothelial and endothelial nature respectively<sup>12</sup>.

In our case the lining cells were neither in continuity with the serosa nor with the tubal epithelium as it happens in most of the cases<sup>1</sup>.

The interstitial tissue surrounding the individual tubules of the tumour showed lymphocytes and plasma cells. This finding has been observed by many authors and even lymphoid follicle formation in the stroma has been documented<sup>2,3,8,10</sup>.

The free margins of the cells were defined, whereas the other cell borders were indistinct, forming a syncytium, in coherence with the observation of Jackson<sup>24</sup>.

Periodic acid– Schiff (PAS) and Alcian blue positive, hyaluronidase digestible material is proven to be present in the lumina of the tubules, cells and intercellular spaces<sup>1,11,19</sup>. However it is said to be present in frozen sections only; lost during routine processing and is not seen in permanent sections<sup>1</sup>. In our case, we neither found any such material in H & E section, nor could we demonstrate it by any means, owing to the small size of the tumour.

## Conclusion

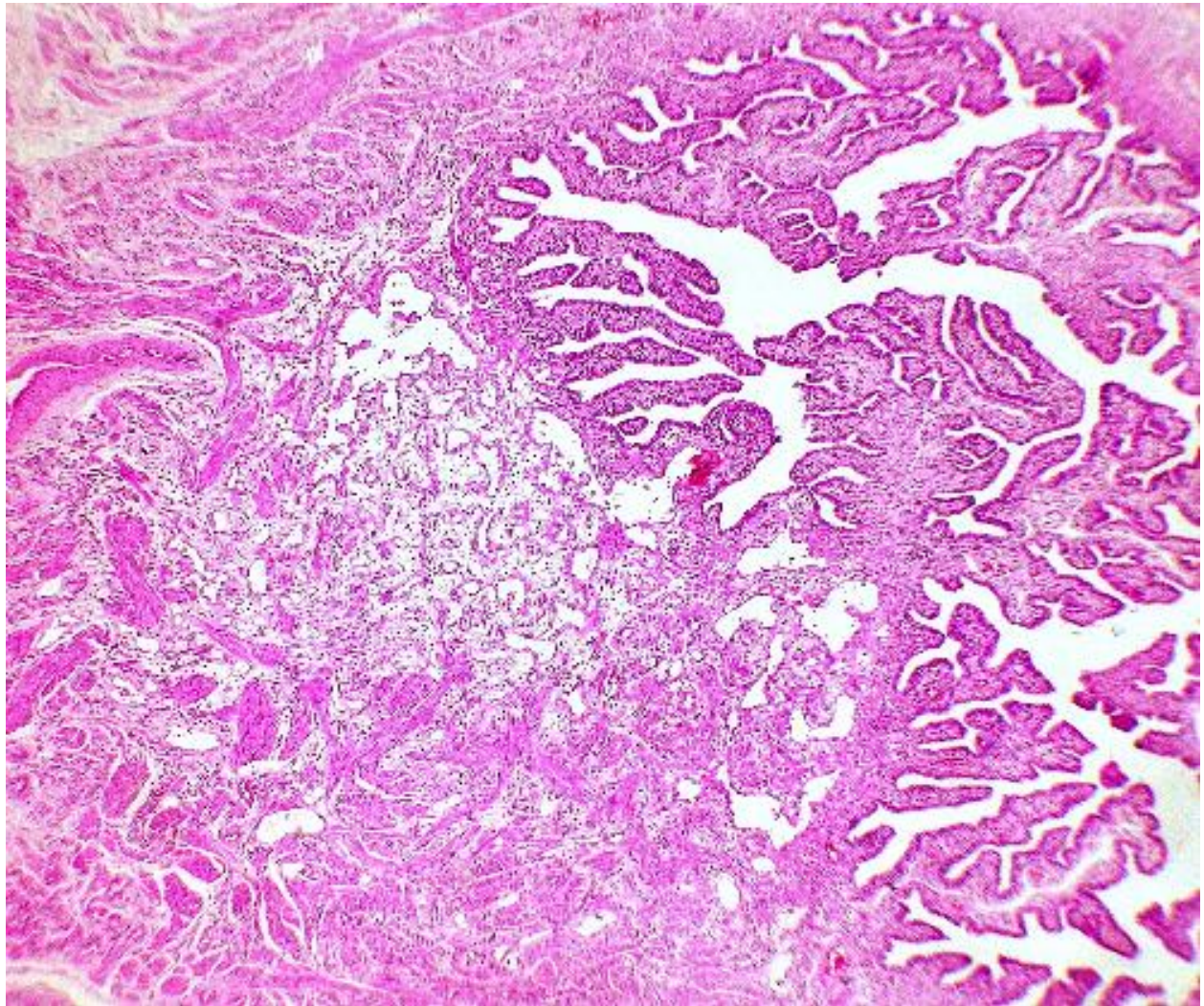
The tumour which we report in this case measured 1 mm across, and is the smallest adenomatoid tumour of fallopian tube ever reported. As reported in the literature, we could not demonstrate any continuity between the tumour cells and the serosa or the mucosal epithelium.

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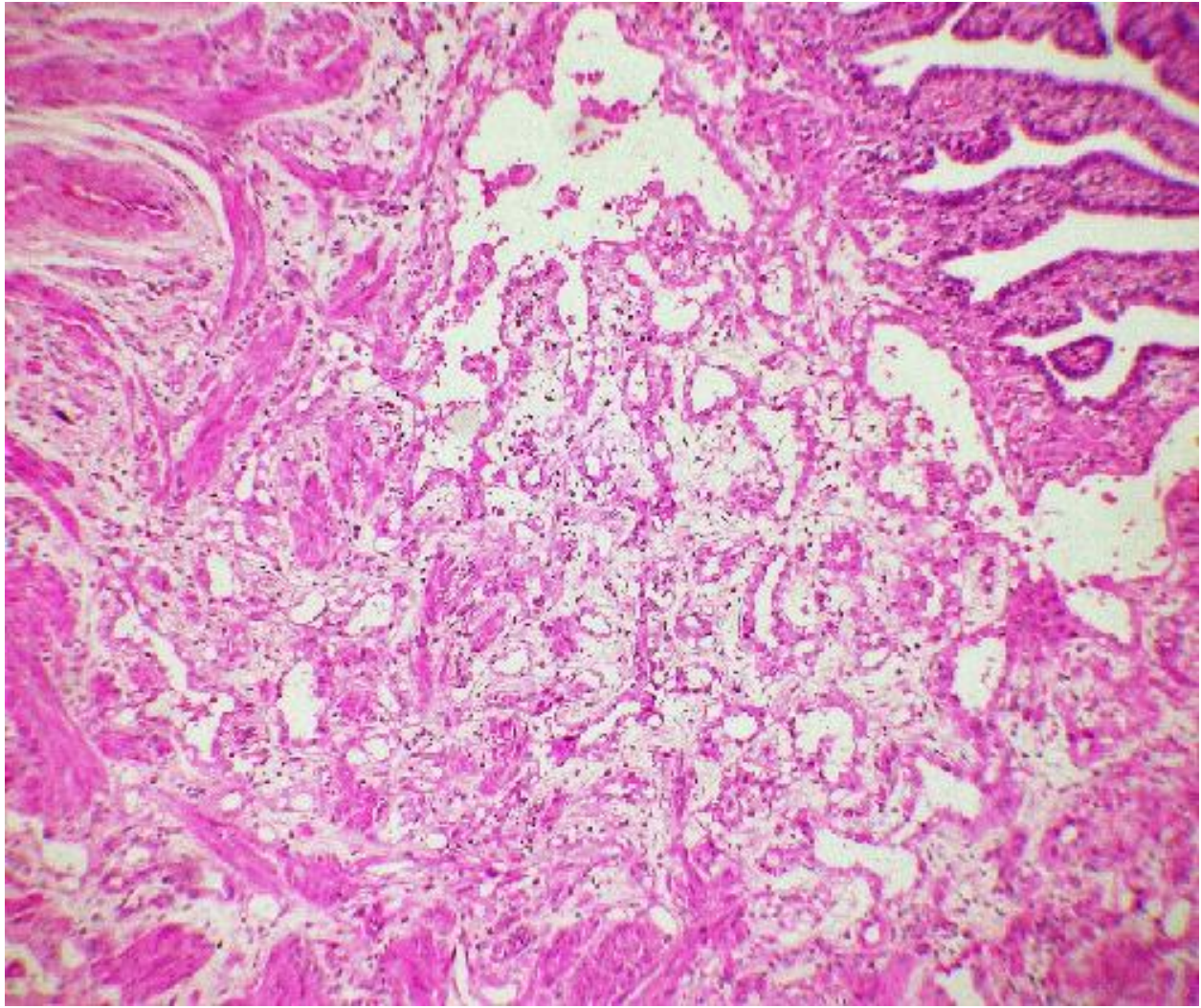
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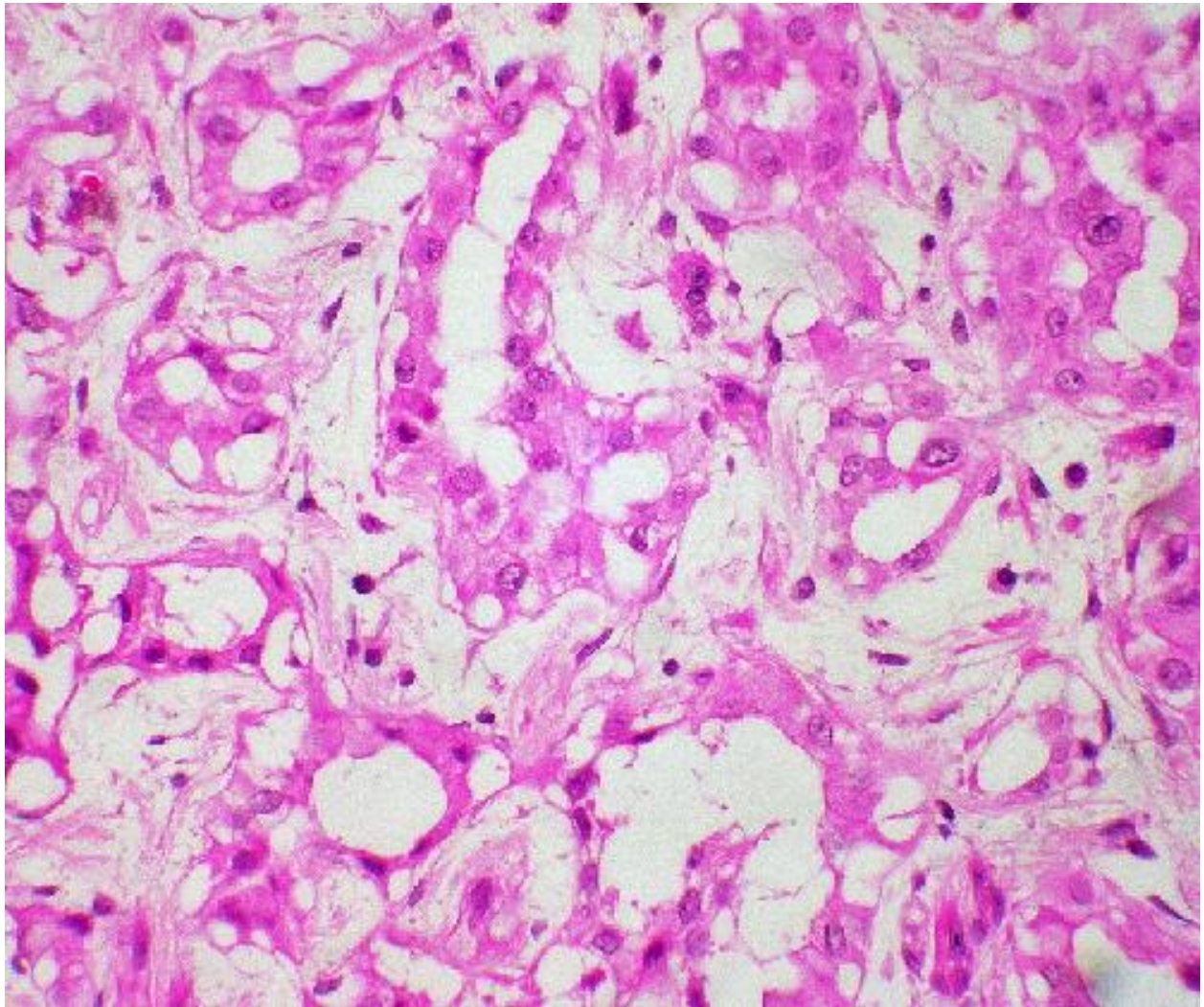
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**Fig. 1:** Circumscribed tumour in the fallopian tube (x 100, H & E)



**Fig. 2:** Tumour located between epithelial and muscular layers (x200, H & E)



**Fig. 3:** Tumour composed of tubules lined by Cuboidal to low columnar flattened cells (x400, H & E)