Sudden Death Associated With Meigs Syndrome
An Autopsy Case Report

Keven K. Hlaise, MBChB, Dip for Med, FC for Path and Sydney M. Shingange, MBChB, Dip for Med

CASE REPORT

Case History
A middle aged woman, in her 40s, was found dead by her teenage daughter in her bedroom on a Sunday afternoon after her return from a morning church service. Her daughter gave a history of stress-related complaints and nothing more. Her last hospital admission was 7 years before her death, when she had her last child delivered by caesarian section.

Autopsy Findings
At autopsy, a 20 × 15 × 18 cm oval-shaped, solid left ovarian tumor with irregular and bumpy surface, and focal cystic areas without normal parenchyma was found (Fig. 1). The right ovary was unremarkable. Approximately 500 mL of straw-colored ascitic fluid was found in peritoneum, approximately 900 mL of straw-colored pleural effusion was found in left chest cavity, and similar and equal amount in right chest cavity. Both lungs were significantly collapsed. The rest of internal organs were unremarkable. She had no cachexia.

Histologic Findings
Histology of the left ovarian tumor showed distorted parenchyma comprising widespread fibrous tissue with well-differentiated fibroblast cells and more or less scanty collagenous connective tissue interspersed between the fibrous tissue without any ectopic tissue (Fig. 2). Other organs were histologically unremarkable.

Cause of Death
In this case, collapse of lungs is the “terminal cause (mechanism) of death,” and the “primary cause of death” is most probably Meigs Syndrome because 3 diagnostic criteria are fulfilled (ovarian fibroma, pleural effusion, and ascites) except there was no way to demonstrate the fourth criteria, ie, resolution of fluid after resection of tumor because the case was diagnosed at autopsy. Meigs syndrome is a natural cause of death.

DISCUSSION
According to Lurie, the syndrome was described in its incomplete form by Spiegelberg in 1888, by Cullingworth in 1879, by Demons in 1887, by Pascale in 1888, and by Terrie in 1888; and in its complete form by Tait in 1892, by Demons in 1900, by Hoon in 1923, by Leo in 1926, by Salmon in 1934, and finally by Meigs in 1934. Hammouda and Lurie separately reported that the description of an association between an ovarian tumor and hydrothorax was first noted by Spiegelberg. Cullingworth was the first to report a case of ovarian fibroma associated with ascites and hydrothorax. Pascale described ascites and hydrothorax in 3 of 21 cases of ovarian cysts. Meigs reported 3 cases in 1934 which were also included in the 7 cases reported by Meigs and Cass in 1937, but one of these 7 cases was that recorded by Dr RC Cabot in 1912. Hammouda mentioned the fact that Rhoads and Terrell, in 1937, designated the syndrome after Meigs, but Meigs was of opinion that this condition be called Demons-Meigs Syndrome since Demons...
reported cases in 1887, 1902 and 1903. Meigs syndrome, although named after Meigs, was first described by Demons of France and Lawson Tait of England, according to Chaitali et al. Stein and Elson noted that although Meigs was the first to recognize and publish the characteristic findings of this condition in 1937, similar isolated cases had already been reported half a century before him. In 1939, Meigs reported 15 cases of Meigs syndrome with similar clinicopathologic presentation; all recovered completely after excision of the ovarian tumor except one patient who died before the tumor could be removed. In 1967, according to Hammouda, there were 129 cases of Meigs Syndrome in the literature. We found one case report in the literature written by South African based authors, ie, R.L. Cheifitz and G. Sher from Department of Obstetrics and Gynecology at Groote Schuur Hospital and University of Cape Town, published in 1976.

The diagnostic criteria postulated by Meigs for diagnosis are: (i) presence of a benign solid ovarian tumor having the gross appearance of a fibroma; (ii) the presence of ascites and hydrothorax; (iii) the disappearance of the fluid, with no recurrence, after removal of the tumor. Stein and Elson quoted Meigs having said in 1939 that: “It is possible, therefore, that there are other benign tumorous conditions in the pelvis besides fibromas of the ovary which may be accompanied by ascites and hydrothorax.” Meigs syndrome embraces any benign tumor of the pelvis which may give rise to ascites and hydrothorax. Lurie re-emphasized the fact that the syndrome is usually associated with fibroma, thecoma, or Brenner’s tumor, and it was also described in association with granulosa-cell tumor, teratoma (specifically struma type). Rapid resolution of fluid accumulation is observed after tumor resection.

Ohsawa et al described Pseudo-Meigs syndrome to be a condition characterized by ascites, and hydrothorax, cured by removing an ovarian or pelvic tumor except ovarian fibroma. The ovarian or pelvic conditions involved are benign tumors of fallopian tube or uterus, mature teratomas, struma ovarii, ovarian leiomyomas, ovarian or metastatic gastrointestinal malignancies, systemic lupus erythematosus, and enlarged ovaries. According to Ohsawa et al, the separation and distinction of true Meigs syndrome from Pseudo-Meigs syndrome is largely academic and does not affect therapeutic aspects of the problem.

Lessnau et al described the pathophysiology of ascites as unknown but emphasized the fact that there were 2 proposed mechanisms; one mechanism that is suggested is the secretion of fluid by tumor and the second mechanism is the direct pressure on surrounding lymphatics or vessels by tumor, hormonal stimulation, and tumor torsion resulting in increased capillary permeability. The cause of pleural effusion is currently unknown but it is theorized that it must be ascitic fluid transferred via transdiaphragmatic lymphatic channels. Characteristically ascites and hydrothorax resolve spontaneously and permanently after removal of the tumor.

Chaitali et al reported that fibromas are the most common tumors of ovarian stroma and constitute 3% to 5% of all ovarian neoplasms. About 91.4% of ovarian tumors associated with Meigs syndrome are fibromas, according to Lanitis et al. About 40% of fibromas greater than 6 cm are associated with ascites. Lessnau et al reported that less than 5% of fibromas are malignant. Meigs, in cases which he described in 1939, reported that the fibromas in his cases varied in size from 9 cm to well over 20 cm. Macroscopically, fibromas are fibrous solid growths that show hard, gray to white whorled cut surface and occasionally focal cystic areas. Meigs thought that cystic areas represent either areas of liquefaction following interference with the blood supply or follicle cysts that have grown large and have not been obliterated. In the cases described by Meigs in 1939, the ascitic and pleural fluid varied from small to very

![FIGURE 1. Ovarian fibroma.](image1)

![FIGURE 2. H&E tissue section of the fibroma.](image2)
large amounts, and its color ranged from the usual straw color to serosanguineous. Histologically, fibromas are composed of bundles of bland spindle cells with elongated nuclei, intersected by bands of collagenous fibrous tissue. Meigs, and Gould and Kerr separately described the ascitic and pleural fluid as transudate, contain lymphocytes but no tumor cells.

Our case is probably the first case report of sudden death associated with Meigs syndrome. In 1879, Cullingworth of Manchester described a case of a woman with Meigs Syndrome who died, without having been operated, from what seemed to be respiratory collapse. In the 15 cases reported by Meigs in 1939, one patient died, before she could be operated, of severe dyspnea and general collapse. Our case has confirmed that untreated Meigs Syndrome is fatal and that the common mechanism of death is lung collapse due to large pleural effusion. Knight, in his book, mentioned that the commonest gynecologic causes of sudden death are ruptured ectopic pregnancy and induced abortions, unless under reputable medical control, from hemorrhage, air embolism, perforation of vagina or uterus, infection and use of toxic substances. We did not find any publication reporting an association between sudden death and Meigs syndrome. The finding and investigation of hydrothorax and ascites in postmortem examinations of females must exclude ovarian tumors. The disturbing questions are: Is it possible that the deceased in our case died suddenly without symptoms, or died without seeking medical help, or the clinicians missed the condition if she ever sought medical help?

ACKNOWLEDGMENTS
The authors thank Mrs Mercia Wilken for her enthusiastic technical assistance.

REFERENCES