# STRUMA OVARII

## (A Case Report with Review of Literature)

by

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*Introduction*

Struma ovarii is a example of unique and rather rare ovarian neoplasm. Although it is not uncommon to note fragments of thyroid tissue in benign cystic teratoma (Dermoids), it is rare to encounter an ovarian tumor composed largely of such tissue (Gusberg and Dan forth, 1944; Smith, 1946; Wynne *et al*, 1940). The term "Struma Ovarii" is asigned to the ovarian neoplasm in which thyroid tissue is the predominant constitutent. Boettlin, 1889, was first to describe the presence of thyroid tissue in a benign cystic teratoma of the ovary. The first published cases of true struma ovarii were by Van Kahlden in 1895 and Gottschalk in 1899, both of whom considered the tumor to be derived from the ovarian follicles. Gottschalk (1899) noted a malignant area in one section of the tumor and therefore designated the neoplasm "folliculoma malignum". Pick, in 1901, was probably the first to appraise the relationship of thyroid tissue to dermoid tumors of the ovary. He concluded that tumors containing thyroid tissue were teratomatous, and he assigned the appellation "teratoma strumoides thyroideale ovarii" to such lesions. The purpose of this paper is to report a case of struma ovarii treated at Medical College Hospital, Aurangabad, and to review briefly the subject.

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### CASE REPORT

Z.S., a 55 years old female was admitted in Medical College Hospital, Aurangabad with the complaints of repeated spontaneous fractures. First fracture was of rib 8 months back. Second fracture was again of rib, 3 months after the first. The third was colle's fracture 8 days before the admission to the hospital. While investigating the case for the cause of these fractures she was submitted for gynaecological examination. On vaginal examination a freely mobile mass was palpable. Abdominal examination also revealed a freely mobile mass in the pelvis.

She was operated 10 years back for multiple fibroids and abdominal total hysterectomy with unilateral salpingo-oophorectomy was performed. The other ovary was normal at that time. She had 5 full term normal deliveries and that delivery was 20 years back.

Laboratory investigation showed serum calcium 9.5 mg.%, serum phosphorus—3.3 mg.%, alkaline phosphatase—8.5 K.A. Units, serum creatinine—1.2 mg.%, blood urea—30 mg.%, serum cholesterol—300 mg%, fasting blood sugar 89 mg.%, postprandial sugar 95 mg.%, urinary calcium 34.77 mgs./24 hrs. urine and urine creatine was 9.5 mg.% Urine Benz Jones protein negative. Electophoresis was negative for 'M' component. Routine haemogram and urinalysis was normal. X-ray examination showed evidence of osteoporosis, no evidence of metastases. A clinical diagnosis of? ovarian tumour with osteoporosis, was made and a laparotomy was done. On laparotomy, a large mass of 5" X 4" was found arising from the ovary which was cystic, lobulated, and well encapsulated, not adherant to surrounding tissue.

**Histopathology**

**Gross:** A large mass of 5" X 4" cystic lobulated, well encapsulated. Cut section showed multicystic tumor (Fig. 1) filled with moderately viscus material. Septae were thickened and many small to large solid areas were seen intervening cystic spaces.

**Microscopy:** Section from the cyst wall showed a thick collagenous band of connective tissue, no lining epithelium was observed. Multiple sections from solid areas and thick septae showed predominantly thyroid tissue. There were acini of various sizes lined by a single layer of flattened or cuboidal epithelium and containing colloid of varying viscosity (Fig. 2). No other teratomatous elements were found on serial sections. There was no evidence of malignancy in all the sections studied.

A histological diagnosis of struma ovarii was given. The patient's postoperative course was uneventful.

*Discussion*

Struma ovarii is composed of true thyroid tissue resulting from unilateral development of an ovarian teratoma. The thyroid tissue present in these ovarian neoplasms is chemically, pharmacologically, biologically, and microscopically identical to cervical thyroid tissue (Plaut, 1933; Stanbury, 1965). The presence of thyroid tissue in a dermoid cyst does not by itself warrant a diagnosis of struma ovarii because such a finding is not uncommon, as evidenced by the 13 per cent incidence quoted by Blackwell *et al*, (1946) in the study of 225 dermoid cysts. On the other hand, tumors composed largely of thyroid tissue and/or those demonstrating physiologic activity or pathologic aberrations are uncommon and tumors fulfilling one of these criteria may be included as examples of "Struma Ovarii" (Wooruff *et al*, 1966).

Smith, in 1946, collected 152 cases of struma ovarii from the world literature. Brocq and associates (1959) determined that 233 cases had been reported until 1959. In 1961, Marcus and Marcus reported 7 cases of struma; however, there were no instances of anaplasia or toxic symptoms. Woodruff and Markley in 1957 reported 4 cases in which there was physiologic or pathologic activity and reviewed 16 instances of malignancy in ovarian struma from the literature. Rotton and Tovell (1956) also reviewed reports of 16 such lesions and added a case of their own. There are approximately 275 bonafide reported cases of ovarian struma (Woodruff *et al*, 1966). 25 cases of struma ovarii seen at the Mayo clinic since 1927 were reviewed in detail by Kempers *et al*, (1970) bringing the total upto 300 cases. In the Indian literature very few cases have been reported (Chattoraj, 1963, Pande and Rajvanshi, 1973).

Struma ovarii have been reported in all age groups, including children (Merttens, 1897). In a review of 152 cases in the literature upto 1946. Smith (1946) found an age range of 6 to 74 years with an average age of 42 years. In Kempers *et al*, series (1970) the average age was 48 years with a range of 19 to 70 years. Only 5 of 25 patients were less than 40 years old. This concentration of cases in upper age range is of interest because

dermoids and more complex teratomas are common in the younger age groups. Thompson (1965) found that these tumors comprise 37.7 per cent of ovarian tumors in children. It can be assumed, therefore, that the thyroid tissue in these strumal tumors is very slow growing and that it takes many years of hypertrophy and involution before sufficient bulk is achieved to make these tumors symptomatic. In Kempers *et al* (1970) series, in one of the asymptomatic cases the pelvic tumor was known to have enlarged over a period of 22 years.

A fascinating aspect has been the occasional occurrence of thyrotoxic signs and symptoms associated with struma ovarii (Brandenberg, 1936; Emge, 1940; Gusberg and Danforth, 1944; Kleine, 1934; Kempers *et al*, 1970; Kovacs, 1924; Marcus and Marcus, 1961; Mc Garrity and Dodson, 1948; Masson and Mueller, 1933; Moench, 1929; Neumann, 1937; Smith, 1946). In certain cases this relationship was not recognised until after regression of symptoms following removal of ovarian tumors (McGarrity and Dodson, 1948; Moench, 1929). In other patients, removal of the ovarian struma resulted in enlargement of the thyroid gland (Foulkes and Fraser, 1954; Woodruff and Markley, 1957) or in actual appearance of symptoms of hyperthyroidism. In Smith's collected series (1946), cervical goiter was noted in 16 per cent of 153 cases. It has been estimated that 5-6 per cent of ovarian strumas produce thyrotoxicosis. The present case was purely asymtomatic and presence of struma ovarii was an incidental finding.

The tumor is usually unilateral. In the present case as one ovary was removed 10 years back together with the uterus and fallopian tube for multiple fibroids and there was no evidence of any pathology in that, the case reported remains as unilateral involvement. Smith (1946) noted thyroid tumor bilaterally in only 4 per cent of 153 cases. The size of the tumor in the literature varied from 5 cm. to 20 cm. (Smith, 1946; Masson and Muller 1933) to a size of child's head (Gottschalk, 1889; Marcus and Marcus, 1961). There was no evidence of spread beyond the ovary. On the basis of the data in this case, the struma was considered benign. Rare instances of adenomatous proliferation (McGarrity and Dodson, 1948; Plaut, 1933; Woodruff and Markley, 1957), papillary tumors (Manasse, 1926; Morgen, 1924) and adenocarcinomas (Moench, 1924, Wynne *et al*, 1940) have been reported in the literature. On the basis of reports to date, it appears that about 5 to 10 per cent of ovarian strumas are malignant. In rare cases the local implants may be benign (Emge 1940). Metastasis to the omental and mesenteric nodes have been reported by Emge, (1940); Shapiro, (1930); and Werth, (1928), to the liver by Proescher and Reddy (1910), to the chest by Woodruff and Markley (1957)., and to bone by Marcus and Marcus (1961). It is interesting that a bone metastasis may be the first sign of the disease and also that symptoms of hyperparathyroidism may subside following excision of the bone lesion. There was no evidence of bone lesion in the present case; though there were multiple fracture suspected due to metastasis X-ray examination revealed fractures were due to senile osteoporesis and there was no evidence of metastasis. The case of metastasis to the chest reported by Woodruff and Markely (1957) is noteworthy since the metastasis occurred 5 years after removal of the ovarian tumor.

One of the most perplexing features of this neoplasm is its histogenesis. Von

Kahlden in 1895 and Gottschalk in 1899 considered the tumor to be derived from the ovarian follicles. Kretschmar (1901) believed the tumor to be an endothelioma. Pick (1901) postulated its origin from dermoids or teratomas. Bell (1905), contended that this tumor resulted from degenerative changes in a pseudomucinous cystadenoma, while Bauer (1914), believed that this lesion was really a cystademoma of an ovary resulting from downgrowths of the surface epithelium in thyroid like arrangement. While it is true that struma ovarii is occasionally found associated with pseudomucinous cystadenoma, there is now general agreement that this tumor is composed of genuine thyroid tissue and represented unilateral development in an ovarian teratoma. A coexistance of Brenner tumor and struma ovarii has been reported (Klein *et al,* 1968).

*Summary*

Struma ovarii a rare benign asymptomatic unilateral ovarian tumor is reported and the literature is reviewed briefly.

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*See Figs. on Art Paper VI*