A Rare Case of Virilizing Ovarian Fibrothecoma in A Teenage Girl

*Ansari NP¹, Biswas JP², Sen S³, Abrar S⁴, Spriha FR⁵

Abstract

Ovarian fibrothecomas comprise tumors in the spectrum of ovarian sex cord / stromal tumors where there are components of both an ovarian fibroma and an ovarian thecoma. They account for 3-4% of all ovarian tumors. Most occur in adult women, with 65% in postmenopausal women. However, it represents only 2% of pediatric ovarian tumors. They are the most common benign solid ovarian tumor. The tumors are usually hormonally inactive but can be estrogenic or sometimes androgenic. Here we are reporting a rare case of a 15-year-old girl presenting with primary amenorrhea, severe hirsutism and finally diagnosed with ovarian fibrothecoma. After surgical removal of the tumor, a dramatic response was observed as the patient developed menarche. She was relieved of her hirsutism by laser therapy later, but her deep voice remained the same.

CBMJ 2024 January: vol. 13 no. 01 P: 95-97

Keywords: Hirsutism, amenorrhea, benign ovarian tumor, teenage girl

Introduction

Sex cord stromal tumors represent approximately 8% of all ovarian neoplasms.¹ Tumors arising from the ovarian stroma are composed of either fibroblasts (fibroma) or plump spindle cells with lipid droplets (thecoma) and many tumors contain a mixture of these cells and called fibrothecoma are relatively common and accounts for 3-4% of all ovarian tumors.² The mean age of occurrence is 45 to 55 years.^{3,4} Pure thecoma are rare, but tumors in which these cells are predominant are hormonally active.²

Case Summary

A 15-year-old unmarried girl came to a specialist dermatologist with the presenting complaints of masculinizing features of severe hirsutism and deep voice along with primary amenorrhea and underdeveloped breast. Investigation through ultrasonogram of lower abdomen revealed a left ovarian solid mass. The uterus and cervix were unremarkable. Hormones study showed serum testosterone level of 228.62 ng/dl, TSH was 2.12 mIU/ml, prolactin was 15.30 ng/ml, and

estradiol/estrogen was 37.62 pg/ml. Moreover, anti-mullerian hormone was found to be 1.29 ng/ml, FSH was 0.97IU/L and LH was 0.35 IU/L. Then the patient underwent surgical operation, and the mass was removed. Histopathological examination of the excised specimen was done. After one week of operation, the patients developed menarche and post-operative follow up revealed a decreased testosterone level.

- *Dr. Nazma Parvin Ansari, Associate Professor & Head, Department of Pathology, Community Based Medical College, Bangladesh (CBMC, B), Mymensingh.
- 2. Dr. Joy Prokas Biswas, Associate Professor & Head, Department of Pathology, Netrokona Medical College, Netrokona.
- Dr. Swagota Sen, FCPS (Dermatology) Part-2 Trainee, CMH, Dhaka.
- 4. Dr. Shakib Abrar, Medical Officer, Upazila Health Complex, Daudkandi, Cumilla.
- Fabiha Rahman Spriha, 5th Year MBBS Student, Dhaka Medical College, Dhaka.

Address of Correspondence:

Email: palashansaripath@gmail.com



Fig. 1: Severe hirsutism was observed in the patient.



Fig. 2: Surgical removal of the tumor.



Fig. 3: Cut sections of the tumour show solid whitish and yellowish surface.

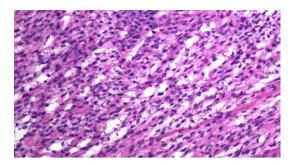


Fig. 4: Histopathological examination showed a benign tumor composed of an admixture of fibrous and thecomatous elements.

Discussion

Ovarian thecoma-fibroma is one of the benign variants of sex cord stromal tumours which affect women infrequently.^{2,5,6} However, its clinical presentation is variable and non-specific.7 It has a wide range of clinical manifestations due to various factors but can also be asymptomatic.8 with Some present amenorrhea/ may oligomenorrhea slowly and progressing virilization. 9,10 It may be presented with features like hirsutism and a deep voice. Our patient had primary amenorrhea, hirsutism, and deep voice. This case highlights that fibrothecoma is the possibility of the menstrual irregularities in the young women. The patient was only 15 years old, younger than usual, presented with hirsutism and primary amenorrhea and lower abdominal lump. Patients with ovarian thecoma-fibroma who have disorder demonstrate menstrual may derangement in hormone profile, 5,9,10 as same as observed in our patient. The treatment is usually individualized but surgical removal of ovarian mass, histopathology and follow up have been commonly reported.²⁻⁴ Surgical removal of the solid ovarian tumor is recommended for proper diagnosis, curative purpose and to reduce the

probability of malignancy and recurrence. Diagnosis is to be confirmed by histopathology. ^{2,3} Soon after surgery, the patient had menarche. She was relieved of her hirsutism by laser therapy later, but her deep voice remained the same.

Conclusion

Fibro-thecoma is a benign tumor of the ovary, which is very rare in young patients and an important cause of amenorrhea and as well as masculinizing features. A careful clinical evaluation, surgical operation (excision) followed by histopathological diagnosis are recommended.

References

- Tavassoli FA, Mooney E, Gersell DJ, et al. Sex cord-stromal tumours. In: Tavassoli FA, Devilee P. eds. Pathology and Genetics of Tumours of the Breast and Female Genital Organs. WHO Classification of Tumours, 3rd ed. Lyon, France: IARC Press: 2003.
- 2. Young RH. Sex cord-stromal, steroid cell, and other ovarian tumors with endocrine, paraendocrine, and paraneoplastic manifestations. In: Kurman RJ, Ellenson LH, Ronnett BM, eds. Blaustein's pathology of the female genital tract. 6th ed. London, UK: Springer; 2011.
- Chechia A, Attia L, Temime RB, Makhlouf T, Koubaa A. Incidence, clinical analysis, and management of ovarian fibromas and fibrothecomas. Am J Obstet Gynecol. 2008;199(5):473.e1-4.
- Cho YJ, Lee HS, Kim JM, Joo KY, Kim ML.
 Clinical characteristics and surgical management options for ovarian fibroma/

- fibrothecoma: a study of 97 cases. Gynecol Obstet Invest. 2013;76(3):182-7.
- Horta M, Cunha TM. Sex cord-stromal tumors of the ovary: a comprehensive review and update for radiologists. Diagn Interv Radiol. 2015;21(4):277-86.
- Haroon S, Zia A, Idrees R, Memon A, Fatima S, Kayani N. Clinicopathological spectrum of ovarian sex cord-stromal tumors; 20 years' retrospective study in a developing country. J Ovarian Res. 2013;6(1):87.
- Meyer AC, Papadimitriou JC, Silverberg SG, Sharara FI. Secondary amenorrhea and infertility caused by an inhibin-B-producing ovarian fibrothecoma. Fertil Steril. 2000;73(2):258-60.
- Mawad NM, Hassanein OM. Ovarian fibrothecoma in a 19 years old Sudanese girl. Gynaecological case report. Clin Exp Obstet Gynecol. 1994;21(4):243-5.
- McGee J, Fleming NA, Senterman M, Black AY.
 Virilizing luteinized thecoma of the ovary in a 15-year-old female: a case report. J Pediatr Adolesc Gynecol. 2009;22(5):e107-10.
- 10. Gaspari L, Paris F, Taourel P, Soyer-Gobillard MO, Kalfa N, Sultan C. Adolescent ovarian thecoma presenting as progressive hyperandrogenism: case report and review of the literature. Gynecol Endocrinol. 2020;36(9):839-42.