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Case Report

Incidentally Detected Ectopic Adrenal Cortical Rest in Ovarian Hilum, a Case Report

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Abstract

It has been proposed by various authors that ectopic adrenal tissue can be identified in approximately 50% of newborns. However, most ectopic adrenal tissues become atrophic with age. These are usually asymptomatic. However in some cases endocrine symptoms and neoplastic transformations have also been reported. As a consequence, it has been suggested that ectopic adrenal tissues should be excised if incidentally encountered during surgery. We here report a case of incidentally detected ectopic adrenal rest located in ovarian hilum in a 44 years old female.

Keywords: Ectopic adrenal rests, incidental adrenal ectopia, ovary, female.

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INTRODUCTION

The first case of ectopic adrenal tissue described by Morgagni in 1740. Many cases have been reported involving various sites in the abdominopelvic cavity, most frequently associated with urogenital structures (Schechter, 1968). The known areas for ectopic adrenal tissue include kidney, liver, pancreas, colon, celiac plexus, testis, broad ligament, placenta, ovary and retroperitoneal area. Ectopic adrenal tissue is more common in male than female genital organs, particularly during childhood, and the most common site is the spermatic cord (Mendez *et al.*, 2006). It has been proposed by various authors that ectopic adrenal tissue can be identified in approximately 50% of newborns. However, most ectopic adrenal tissues become atrophic with age (Souverijns *et al.*, 2000).

CASE REPORT

We present here a case of a 44 years female who presented with severe pain in abdomen since a week. The ultrasonography revealed a 2.2 cms sized haemorrhagic right ovarian cyst. A laparoscopic salpingo-oophorectomy was performed. We received multiple congested grey brown tissue pieces aggregating to 3 x 2 x 2 cms. On cut surface few of the tissue pieces were solid grey white while others had appearance of a cyst wall with yellowish brown appearance. Few blood clots were present. Ovarian hilar adipose tissue revealed a well circumscribed encapsulated nodule measuring 5 x 2 x 2 mms. The cut surface of nodule appeared yellowish. On microscopic

examination, this nodule revealed ectopic adrenal gland rest. It was comprising cortical adrenal cells only devoid of the adrenal medulla. Resembling the normal adrenal gland, this rest revealed a fibrocollagenous capsule adjacent to which was thin zona glomerulosa, followed by thick layer of zona fasciculata. Innermost was thick layer of zona reticularis. A haemorrhagic luteal cyst of ovary was identified along with adjacent ovarian tissue containing corpora albicans. The fallopian tube revealed hydrosalpinx.

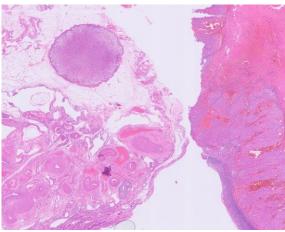


Figure 1: On the right is seen ovary with a haemorrhagic corpus luteal cyst. On the left is seen ovarian hilum containing adipose tissue and vessels. Within this hilar tissue in the upper left corner of picture is seen a well circumscribed encapsulated nodule of ectopic adrenal rest. H&E, 5x

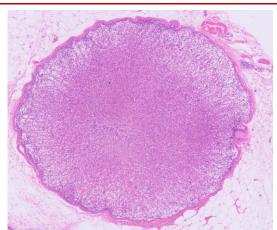


Figure 2: The ectopic rest comprises only the adrenal cortical tissue, no adrenal medullary tissue is seen. H&E, 20x

DISCUSSION

The adrenal glands are of double embryological origin. The adrenal cortex is derived from the coelomic mesoderm of the urogenital ridge, while the adrenal medulla develops from the neural tissue (Barwick et al., 2005). During embryological development medullary tissue invades the cortex along the central vein. Defects in this penetration process may lead to small fragments, which subsequently separate and migrate with descent of the gonads. Ectopic adrenal rests exist along the migration path of adrenal cortex development, and these anatomic sites include the celiac plexus, kidney, ovary, broad ligament, testis, and spermatic cord (Çağdaş et al., 2017). The brain, lungs, and stomach have been reported to be rare sites of ectopic rests. As the adrenal medulla has a different embryologic development that being from the neural crest, these ectopic rests are composed only of the adrenal cortical tissue. Similarly in our case, the ectopic adrenal rest comprised only adrenal cortical tissue and with similar composition to that of the normal adrenal gland. Ectopic adrenal tissue is found in 50% neonates, and most of the ectopic rests undergo atrophy (Tsai et al., 2021). The occurrence of adrenal rest tissue in adults is 1% (Souverijns et al., 2000). The size of these ectopic adrenal rests ranges from 2 to 40 mms, the average size is 7.6 mms. In our case the ectopic adrenal rest measured 5 x 2 x 2 mms in size. These ectopic adrenal tissues become atrophic with age due to normally functioning adrenal glands. These are usually asymptomatic (Senescende et al., 2016). However in some cases endocrine symptoms such as hypertension and fasciotruncal obesity, virilization due to hormonal activity can be seen. Also corticotropin-independent Cushing's Syndrome can arise. Neoplastic transformations of ectopic adrenal tissue like adrenocortical adenoma, carcinoma, myelolipoma has also been reported (Tsai et al., 2021, Dotto et al., 2019, Burman et al., 2021). As a consequence, it has been suggested that ectopic adrenal

tissues should be excised if incidentally encountered during surgery (Mendez *et al.*, 2006).

CONCLUSION

In conclusion, ectopic adrenal tissues rare in adult females. These are mostly asymptomatic and revealed incidentally during surgery. Few may give rise to obesity, hypertension and some cases have demonstrated the risk of neoplastic transformation. Therefore, we must be aware of this rare entity, and surgically remove all incidentally found suspicious lesions.

REFERENCES

- Schechter, D. C. (1968). Aberrant adrenal tissue. *Annals of surgery*, 167(3), 421–426. https://doi.org/10.1097/00000658-196803000-00017
- 2. Mendez, R., Tellado, M. G., Somoza, I., Liras, J., Sanchez-Abuin, A., Pais, E., & Vela, D. (2006). Ectopic adrenal tissue in the spermatic cord in pediatric patients: surgical implications. *International braz j urol*, *32*(2), 202-207.
- 3. Souverijns, G., Peene, P., Keuleers, H., & Vanbockrijck, M. (2000). Ectopic localisation of adrenal cortex. *European Radiology*, *10*(7), 1165-1168.
- Barwick, T. D., Malhotra, A., Webb, J. A. W., Savage, M. O., & Reznek, R. H. (2005). Embryology of the adrenal glands and its relevance to diagnostic imaging. *Clinical radiology*, 60(9), 953-959.
- Şahin, Ç., Taylan, E., Akdemir, A., Zekioglu, O., Seyidova, P., & Ergenoglu, A. M. (2017). Ovarian serous cystadenoma with ectopic adrenal tissue in a 65-year-old patient: A case report. *International* journal of surgery case reports, 33, 89-91.
- 6. Tsai, W. H., Chen, T. C., Dai, S. H., & Zeng, Y. H. (2021). Case Report: Ectopic Adrenocortical Carcinoma in the Ovary. *Frontiers in endocrinology*, *12*, 662377. https://doi.org/10.3389/fendo.2021.662377
- 7. Senescende, L., Bitolog, P. L., Auberger, E., Le Bian, A. Z., & Cesaretti, M. (2016). Adrenal ectopy of adult groin region: a systematic review of an unexpected anatomopathologic diagnosis. *Hernia*, 20(6), 879-885.
- 8. Dotto, R. S., Marx, G., Bastos, M., Machado, J. L., Glufke, V., & de Oliveira Freitas, D. M. (2019). A rare case of virilizing adult ectopic adrenal tumor. *Urology case reports*, 27, 100907.
- Burman, P., Falhammar, H., Waldenström, E., Sundin, A., & Bitzén, U. (2021). 11C-metomidate PET/CT detected multiple ectopic adrenal rest tumors in a woman with congenital adrenal hyperplasia. *The Journal of Clinical Endocrinology* & *Metabolism*, 106(2), e675-e679.