

Pediatrics

Ectopic adrenal tissue in an inguinal hernia sac: A case report

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ARTICLE INFO

Keywords:

Ectopic adrenal tissue
Adrenal tissue in inguinal hernia sac
Case report
Adrenal ectopy

ABSTRACT

Adrenal ectopy is the rare occurrence of adrenal tissue at abnormal locations with hernia sac being particularly infrequent. 2-year-old male child with swelling in right inguinal region was diagnosed as reducible inguinal hernia. Intraoperatively during hernia repair, the sac was identified with a thick yellow nodule adherent to it. Separation of sac from cord structures along with high ligation of sac was done. The histopathological analysis of abnormal tissue was consistent with ectopic adrenal cortical tissue. We conclude that abnormal tissue found incidentally at inguinoscrotal surgeries should be removed, since ectopic adrenal tissue can potentially cause morbidity in the future.

1. Introduction

Ectopic adrenal remnants are nodules of heterotopic adrenocortical tissue, often discovered incidentally during surgical procedures conducted for other reasons. This rare condition was first identified by Morgagni in 1740 in the vicinity of the adrenal gland. The estimated incidence is 1.7 to 3 %¹. It is frequently found in males during inguinoscrotal surgeries, commonly inguinal herniotomy and orchidopexy, with reports of adrenocortical tissue located in the testes or along the spermatic cord. Rare cases of adrenal ectopy have been documented in females, including locations such as the para-ovarian area, fallopian tubes, and broad ligament. The development of adrenal cortical tissue originates from mesoderm that lies medial to the developing gonads. Consequently, foci of this developing cortex usually accompany the gonads as they descend and may get deposited along their path. In contrast, the adrenal medulla arises from the neuroectoderm, making it less likely to be part of this condition.² Clinical implications of ectopic adrenal rest include the potential for malignant transformation, functional hypertrophy of the ectopic tissue in patients who have undergone bilateral adrenalectomy, and acute adrenal insufficiency following the accidental removal of the sole ectopic adrenal tissue in patients who lack normal adrenal glands.

2. Case report

A two-year-old male was brought by his parents due to a right

inguinoscrotal swelling that had been noticed one month before, with no associated symptoms. On examination, a soft swelling was palpable in the right inguinoscrotal region which was completely reducible, with no skin changes or tenderness. It was diagnosed clinically as a right inguinal hernia. The patient was taken to the operating room electively for a right inguinal hernia repair. During the procedure, the hernial sac was carefully dissected off the spermatic cord structures. At this point, an abnormal firm yellow nodule was observed along the hernial sac [Fig. 1]. The hernia sac was ligated proximally at the deep inguinal ring. A section of the hernia sac containing the abnormal nodule was sent for a histopathological analysis. The patient had an uneventful recovery, and he was discharged on the same day. The histopathological examination of the specimen revealed a grey - brown firm nodule along the hernia sac, measuring approximately 1 cm × 0.8 cm × 0.5 cm. The microscopic examination showed a nodule composed of two distinct cell populations, including multiple vacuolated, lipid-rich cells resembling those of the zona fasciculata, admixed with eosinophilic cells like those found in the zona glomerulosa. This confirmed the diagnosis of ectopic adrenal tissue adherent to the inguinal hernia sac [Fig. 2]. On follow up the patient did not develop any clinical signs related to the excision of the ectopic adrenal tissue.

3. Discussion

Accessory adrenal tissue is a rare incidental finding that occur in 0.3 %–0.7 % of inguinal hernia repairs according to the literature.^{3,4} While

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<https://doi.org/10.1016/j.eucr.2025.103145>

Received 9 June 2025; Received in revised form 22 July 2025; Accepted 29 July 2025

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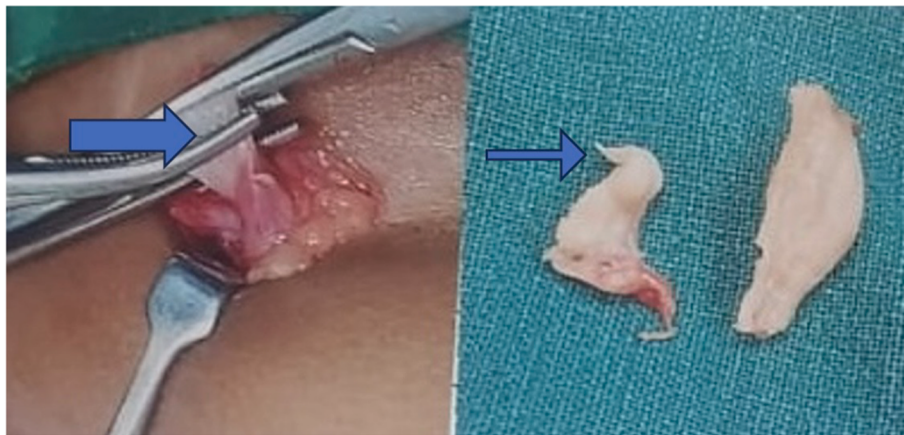


Fig. 1. Intraoperative finding of a thick firm yellow nodule adherent to the hernia sac (arrows).

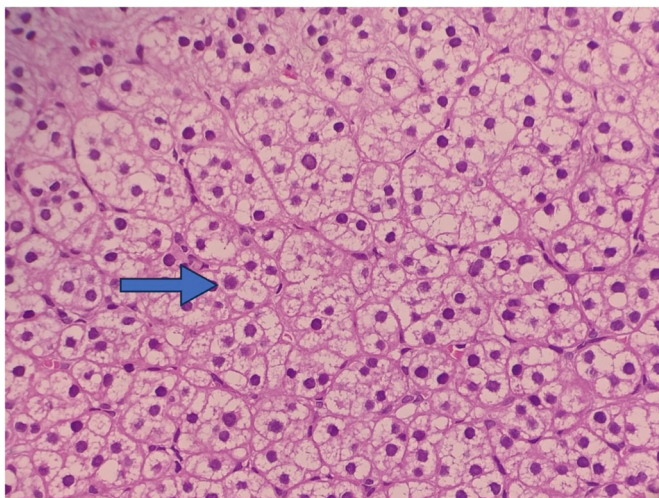


Fig. 2. High-power histology view showing large polygonal cells with distinct cell membranes and eosinophilic cytoplasm (arrow).

common locations for ectopic adrenal tissue include the area around the celiac axis and the kidneys, unusual locations such as the placenta, liver, lungs, and intracranial cavity have also been documented. This supports the theory that accessory nodules can develop in multiple sites from pluripotent cells as well as accompany the gonads during their descent leading to ectopic locations along the migratory pathway.^{5,6} True heterotopia of the adrenal gland occurs when there is failure in separation of the cortex, which develops from coelomic mesothelium at 7–8 weeks of gestation and is associated with incomplete descent of cortical cells and descending gonads.^{7,8} Most cases demonstrate the presence of these benign nodules in retroperitoneal area, which are grossly round and yellow, measuring between 1 and 5 mm in diameter.⁹ Histological examination of these nodules typically exhibits immunohistochemical positivity for melanin A, inhibin, synaptophysin, and calretinin, and high nuclear positivity for the antisteroidogenic factor 1. They stain negative for chromogranin. Most of these nodules are asymptomatic and simply diagnosed incidentally, and in most cases, the tissue is hormonally inactive.¹⁰ However, there are reported cases in which those ectopic nodules of adrenal tissue had clinical implications, such as compensatory functional hypertrophy in patients who had undergone bilateral adrenalectomy for pathological adrenocorticotrophic hormone production, or in individuals with congenital adrenal hyperplasia, which can lead to a recurrence of symptoms.^{11–13} Additionally, there are reports of malignant transformation of ectopic adrenal tissue

into conditions such as adrenocortical carcinoma, pheochromocytoma, Leydig cell tumor, and neuroblastoma, although its presence during inguinal hernia surgeries has not been documented so far.^{14,15} These aberrant cells may mimic a metastatic renal cell carcinoma, carcinosarcoma, or malignant melanoma, highlighting the importance of awareness to avoid misdiagnosis.^{16,17} Theoretically, true adrenal heterotopias may be the only functioning adrenal glands, its removal could cause adrenal insufficiency.¹⁸ Homeostatic endocrine mechanisms may get disrupted if the tissue is hyperfunctional with suppression of hypothalamic-pituitary-adrenal axis. Other anatomically aberrant specimens encountered during hernial sac analysis may include various metastases, endometriosis, bladder tissue, appendices, inclusions from Wolffian or Müllerian remnants, along with mesothelial hyperplasia due to trauma or inflammation.¹⁹ While excision of ectopic tissue is generally considered straightforward, routine intra-operative searches for these structures are discouraged, as they can risk disrupting the native anatomy.²⁰

4. Conclusion

Abnormal tissue found in an inguinal hernia sac should be removed, since ectopic adrenal tissue can potentially cause morbidity in the future, such as malignant degeneration.

CRediT authorship contribution statement

Nandini Sateesh Desai: Writing – original draft. **Aditi Dalvi:** Writing – review & editing, Supervision. **Sanjay Oak:** Writing – review & editing, Validation, Supervision, Data curation, Conceptualization. **Paras Kothari:** Visualization, Supervision. **Abhaya Gupta:** Writing – review & editing, Validation, Supervision, Conceptualization. **Shahaji Deshmukh:** Conceptualization.

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