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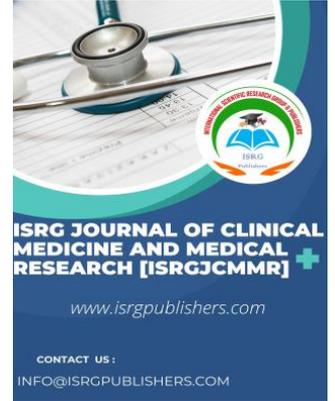
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INCIDENTAL WALTHARD CELL REST WITHIN THE VERMIFORM APPENDIX, an extremely rare entity and literature review

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Abstract

Urothelial rests, which are more commonly known as Walthard cell rests, are usually encountered in the female genital system, while they are very rare in men and outside the genital system. When we look at the literature, appendix localisation is quite rare. Therefore, this lesion should be kept in mind in the differential diagnosis. We shared a 46-year-old male patient with appendiceal localisation with the literature. Our case is the oldest patient according to the literature.

Keywords: *Walthard, Urothelial, Cell, Rest.*

INTRODUCTION

Urothelial rests, also known as Walthard cell rests, are benign incidental findings frequently encountered in the female genital tract (1). Walthard cell rests were first described in 1903 by the Swiss obstetrician Max Walthard in a paper containing 80 series of ovarian sections. Walthard also observed these cell islands on the surface of the fallopian tube and posterior aspect the broad ligament (2,3).

CASE REPORT

A 46-year-old male patient was admitted to the Emergency Department of Eskişehir Yunus Emre State Hospital with complaints of lower quadrant pain for 2 days. On examination, there was tenderness in the lower quadrant and the appendix was found to

be 7 mm on abdominal computed tomography. There were no chronic diseases or complaints in the patient's medical history. Macroscopically, the appendix was 6.8 cm long and 1.1 cm in diameter, with grey-green fibrinopurulent material in the serosa. Microscopically, there was inflammation and serosal fibrinopurulent exudate and mesothelial hyperplasia throughout the appendix wall, including ulceration in large areas of the mucosa (Figure 1), but there was an island of cells with ovoid nuclei and large eosinophilic cytoplasm with visible nuclei in a subserosal area in the apical region of the appendix. The centre of this island of cells was cystic with luminal eosinophilic secretion. Although the entire

material was examined, no other islands of cells were observed in the mucosa and wall (Figure 2).

DISCUSSION

When Walthard cell rests were first described in 1903, only 5 of the patients were observed in the ovarian hilus and were reported to be associated with the surface epithelium in a series of 80 ovaries. While 4 of these patients were under the age of 20 years, 1 of them was 65 years old. In this study, the cells forming these islands were reported to be of 2 types: Group 1 cells were densely packed with dark, spindle-shaped nuclei, and Group 2 cells were round, oval or polygonal cells containing large vesicular nuclei and surrounded by abundant cytoplasm. In this study, it was also stated that they may be cystic. In this study, Walthard stated that he thought that these cell islands were congenitally misplaced (2,3). In the same year (1903), Mayer reported the same cell islands in the fallopian tube, broad ligament, testis and epididymis. He explained this distribution on rather vague embryological grounds and attributed the ability of serosal cells in these regions to form cell nests under inflammatory conditions (2,4). Since then, there have been many discussions about the origin of Walthard cell thicketts. Various theories proposed in this regard include Müllerian remnants, mesothelial metaplasia and metaplastic urothelium (5,6). Today, immunohistochemical staining techniques show a close relationship with urothelial epithelium or urothelial metaplasia (5,7). Walthard cell rests are very rare in the male genital system and appendix compared to the female genital system (1). According to our observation, 6 cases have been described in the literature. Of these 6 cases, 3 were paediatric patients, 2 were adult patients (28-year-old female and 33-year-old male) and one was a male patient and age was not specified. One of the paediatric patients was a 10-year-old female and one was a male (no age information) and gender and age were not specified (1,5,8,9,10,11,12) (Table 1). Our patient was a male patient aged 46 years and appeared to be the oldest patient according to the literature. For differential diagnosis of Walthard cell rests from morphologically confusable lesions such as reactive mesothelial hyperplasia, carcinoid tumour and well-differentiated urothelial carcinoma, it is necessary to know the presence of these lesions (7,10). Immunohistochemical stains are helpful in this regard. Walthard cell rests are positive for CK7, p63, GATA 3 and Invulcrin, while CK20, Uroplakin, Synaptophysin, WT1 and calretinin are negative (1,5,7). It is important to recognise these lesions because they are benign and do not need any other treatment. In conclusion, when we encounter this lesion, which is very rarely encountered in our daily practice, it should be recognised and approached correctly in order to prevent an extra procedure. For this reason, we wanted to share this patient by reviewing the literature.

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Figure 1: Ulceration of the mucosal surface and intense inflammation of the entire wall of the appendix (H&E, X40).

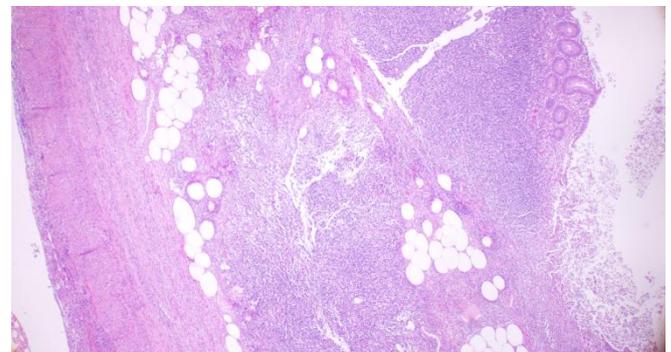


Figure 2: Akut appendicitis with a walthard cell rest within the serosa (H&E, X200);

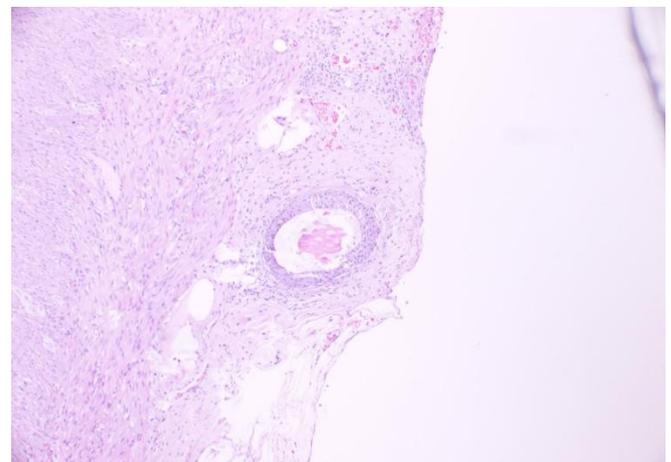


Table 1: The summary of clinic features of previously reported cases

	Years	Age	Gender
Gorter RR, et al.	2017	Pediatric	None
Squires L, et. al.	2018	28 years	Female
Schild MH, et. al.	2019	Pediatric	Male
Punjabi LS, et. al.	2021	33 years	Male
Chang E, et. al.	2022	10 years	Female
Nagalingam S, et. al.	2024	None	Male