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Olgu Sunumu

Adrenal Bölgede İnsidentaloma Gibi Görünen Aksesuar Dalaklı Bir Olgu Sunumu ve Literatürün Gözden Geçirilmesi

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Ö Z E T

Adrenal insidentalomalar sıklıkla adrenal bezle ilgisiz görüntüleme işlemlerinin ardından rastlantısal olarak bulunurlar. Aksesuar dalağa bağlı adrenal insidentaloma hastası güncel endokrin ve radyolojik yönetimle sunuldu.

Elliüç yaşında erkek hasta dispeptik yakınmalar nedeniyle yapılan ultrasonografik incelemede bulunan 25x20 mm boyutlarında sol sürrenal kitle nedeniyle Endokrinoloji ünitesine sevkedildi. Hastanın biyokimyasal ve hormonal incelemeleri normal sınırlardaydı. Bilgisayarlı tomografi ve magnetik rezonans görüntülemeleri ile ayırıcı tanısı yapıldı. Görüntülerinde malignite özellikleri yoktu. Adrenal insidentaloma aksesuar dalakla uyumlu bulundu.

Adrenal insidentaloma gibi görünen aksesuar dalaklı hastaların tanısal değerlendirilmesi klinik ve radyolojik yöntemlerle dikkatle yapılmalıdır.

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Case Report

An Accessory Spleen in Adrenal Region Mimicking as An Incidentaloma: Case Report And Literature Review

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ABSTRACT

Adrenal incidentalomas are often discovered incidentally after an imaging procedure unrelated to the adrenal gland. A patient with an accessory spleen as an adrenal incidentaloma was reported with current endocrine and radiological evaluations.

A 53-year-old man was referred to Endocrinology unit with a mass which is 25x20 mm diameter left surrenal region. This mass is founded in a sonographic examination which was performed for dispeptic complaints. The biochemical and hormonal analyses of the patient were within normal limits. Magnetic resonance imaging was performed to make a definitive diagnosis as the computerized tomography imaging showed no malignant characteristics. The incidentaloma in the left adrenal region has been found to be consistent with an accessory spleen.

Clinical and radiological evaluations of the patients with an accessory spleen in adrenal localization as an incidentaloma should be carefully performed.

1. Introduction

Adrenal masses are often discovered incidentally after an imaging procedure is performed that is unrelated to the adrenal gland and are then termed as adrenal incidentalomas. Its incidence has been increasing proportionally with the use of radiographic imaging (1 – 4). In a patient with a adrenal incidentaloma, several questions arise as to whether the mass is an adrenal or extra-adrenal mass, whether it represents a metastasis of an unknown or known primary tumor, whether the adrenal mass is hormonally active and whether there is evidence of adrenocortical carcinoma. In clinical practice, defining whether lesions in adrenal regions are primary to the adrenal gland or represent other tissues, whether they are benign or malignant and whether they are functioning or not.(5 – 10).

Otherwise, ectopic splenic tissue which known accesory spleen is developed from defective fusion of cells during embriyonic growth. Acceroy spleensnare present approximately 30% of the population, and may be diagnosed as an incidental during radiological imaging, at postmortem, in complicated cases with torsion or rupture (11-14).

In this report, the definitive diagnosis of a patient with accessory spleen detected in adrenal region as an adrenal incidentaloma is pesented.

2. Case Report

In this report, the definitive diagnosis of a patient with accessory spleen detected in adrenal region as an adrenal incidentaloma is pesented.

A 53-year-old man presented with a mass in left adrenal localization about 25x20 mm diameter found in sonographic examinations for dispeptic complaints. He had never smoked and there was nothing remarkable in the family history. On physical examinations, the patient appeared normal, with no cushingoid appearance or dorsal hump. There was no enlarged lymph nodes. His temperature was 36,9˚C, pulse rate was 72/min, respiration rate was 18/min, and blood pressure was 110/70 mm Hg. The lungs and the heart were normal. Abdominal examinations revealed no mass.

Hematologic, blood chemical and hormonal parameters were within the normal range (Table 1, 2 and 3). His electrocardiogram was normal. An examinations by gastroduodenoscopy and upper abdominal sonography because of dyspepsia determined antral gastritis and an incidental mass lesion in the left adrenal compartment.

Table 1. Laboratorial parameters of the case having adrenal incidentaloma

Parameters	Ranges	Results
Fasting glucose (mg/dl)	70 – 100	75
Urea (mg/dl)	10 – 50	36
Creatinine (mg/dl)	0.5 – 1,2	0.7
Hemoglobin (gr/dl)	12 – 18	12.7
Hematocrit (%)	37 – 52	36.7
Mean corpuscular volume (µm³)	80 – 99	84
Erytrocyte sedimentation rate (mm/hr)	< 20	28
White-cell count (per mm³)	4800 – 10.000	6900
Platelet count (per mm³)	130.000 – 400.000	191.000
Total protein (g/dl)	6.4 – 8,3	7.36
Albumin (g/dl)	3.4 – 5,4	3.5
Calcium (mg/dl)	8.4 – 10,2	8.6
Phosphorus (mg/dl)	2.7 – 4,5	2.7
Sodium (mEq/lt)	136 – 157	147
Potassium (mEq/lt)	3.5 – 5,5	4.4
Chloride (mEq/lt)	90 – 110	102
Alanin aminotransferase (U/liter)	10 – 50	12
Aspartat aminotransferase (U/liter)	10 – 40	17
Alkaline phosphatase (U/Liter)	< 129	81

The mass located in the left adrenal region, was oval and measured 25x20cm diameter. Its echogenicity was identical to that of the main spleen, but it was separated from the spleen (Figure 1). The mass was located in the immediate vicinity of the splenic hilum, three were in the region of the splenocolic ligament. Computed tomography and magnetic resonance (MR) imaging was performed to make a definitive diagnosis. Computed tomography images showed that the lesion feature was isodense and similary to spleen (Figure 2). Magnetic resonance images of T1W and T2W axial and coronal planes demonstrated that the accessory spleen was isointense with the main spleen situated in the adrenal compartment (Figure 3 A-C). On in-phase and opposed-phase MR images, the mass was homogeneous with the same signal intensity as the spleen (Figure 3D-E). The conclusions were that the mass in the left adrenal region as an incidentaloma

Table 2. Hormonal analyses of the case

Serum hormones	Ranges	Results
17-α-hydroxyprogesteron (ng/dl)	0.5 – 3.34	1.35
Plasma Renin Activity (ng/ml/hr)	0.2 – 2,8	3.0
Dehydroepiandesterone sulphate (µg/dl)	80 – 560	65.3
Fasting Cortisol (µg/dl)	5 – 25	11.5
Post-dexamethasone suppression cortisol (µg/dl)	< 2	1.5
Adrenocorticotropic hormone (ACTH) (pg/ml)	10 – 46	23
Thyrotropin (TSH) (mU/L)	0.400 – 4.000	0.462
Free T4 (ng/dl)	0.8 – 1,9	1.54

Table 3. The Levels of urinary hormones of the case

Urinary hormones	Ranges	Results
Epinephrine(µg/24 hr)	< 50	0.82
Norepinephrine (mg/24 hr)	< 8	4.23
Metanephrine (µg/24 hr)	< 600	0.82
Normetanephrine (µg/24 hr)	< 600	423
5-hydroxyindoleacetic acid (5-HIAA) (mg/24 hr)	2 – 10	4.2

was consistent with an accessory spleen. This case was admitted with functional dyspepsia and followed up in outpatient clinic with the diagnosis of accessory spleen.

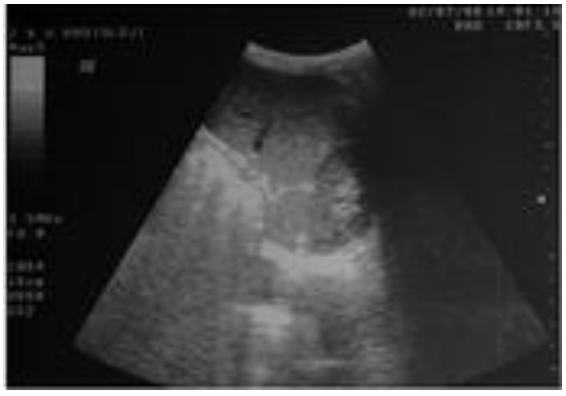

Figure 1. Longitudinal sonogram. Accessory spleen (arrows) separated from main spleen (S) and left kidney (K) by interface.

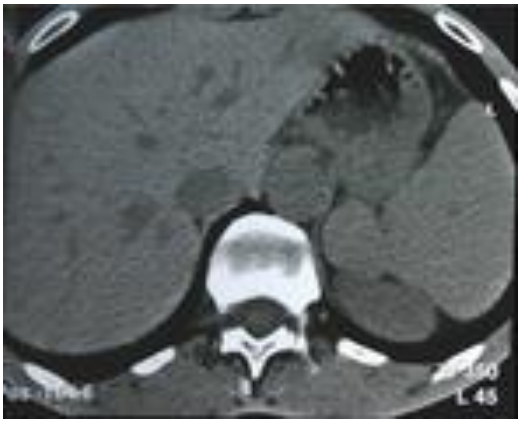

Figure 2. Accessory spleen (arrow) is isodense with main spleen on CT scan.

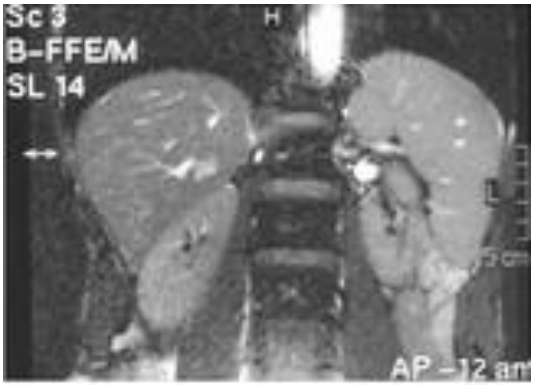

Figure 3A. Coronal T2-weighted turbo spin echo image shows accessory spleen (arrow) is isointense with main spleen situated in surrenal localization (A). Axial T1W

3. Discussion

Accessory spleen may dedected in embriogenic path in the abdomen. It may especially ocur in such areas as the hilum of the spleen, the gastrosplenic and splenorenal ligaments, the tail of the pancreas, the mesentery, the greater omentum, the walls of the stomach or intestines, the adrenals, and the gonads

(12). Satellite splenic tissue can also occur in splenosis due to fragments of splenic tissue at the abdominal cavity after splenectomy or rupture of the spleen (15). There have been reported a few cases with an accessory spleen mimicking adrenal tumour such as adrenal carcinoma, hematologic disoders and solid tumors (12, 16, 17, 18).

Adrenal incidentaloma is a by-product of modern technology, and the routine use of sophisticated radiological techniques has revealed a problem that is not new but is increasingly recognized in current medical practice (5-14, 19-21). Cawood et al. argued that ultrasonography as the primary imaging modality may introduce a selection

bias (21), but in many series published over the past decades that were included in their analysis, adrenal incidentalomas have also been detected by this technique (22-25).

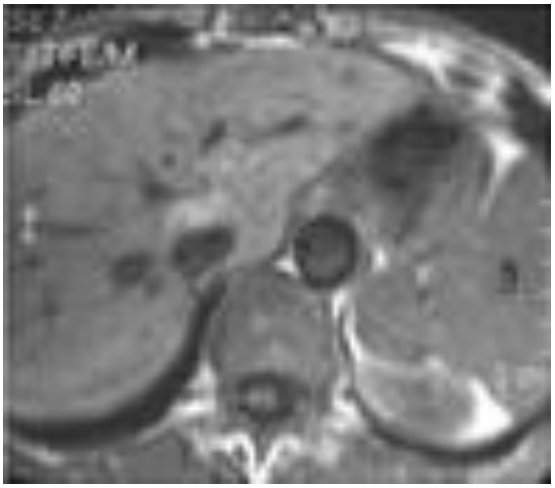

Figure 3B. T2W

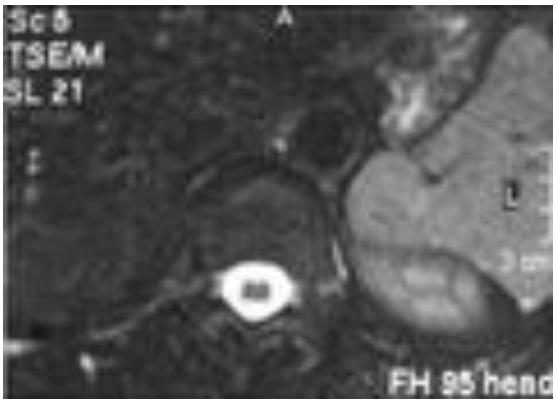

Figure 3C. MR images show accessory spleen (arrow) is isointense with main spleen as an adrenal mass.

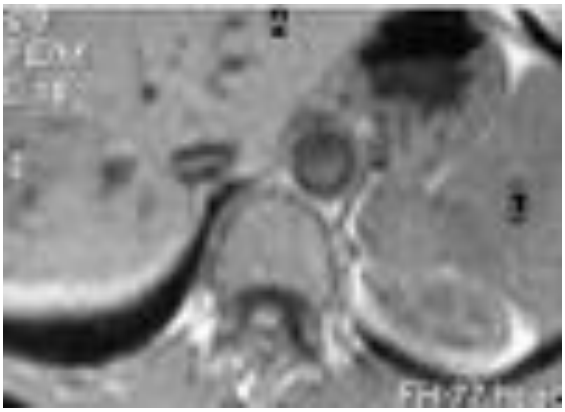

Figure 3D. MR images show accessory spleen (arrow) is isointense with main spleen as an adrenal mass.

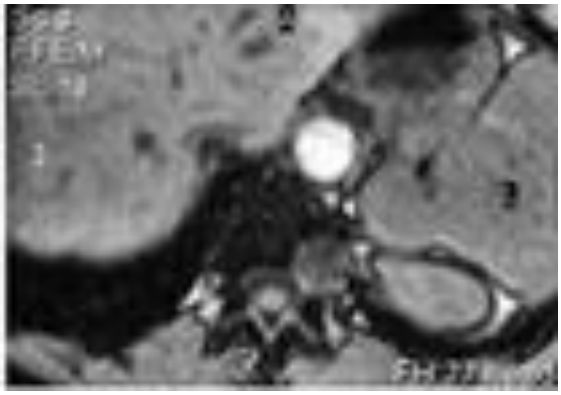

Figure 3E. MR images show accessory spleen (arrow) is isointense with main spleen as an adrenal mass.

Cawood et al. gave a median estimate of around 2,0% for adrenocortical cancer, <1,0% for adrenal metastases and around 3,0% for phaeochromocytoma. Subclinical Cushing's syndrome is the most frequent hormonal disorder among adrenal incidentalomas with a median frequency of about 6,0% (21). An adrenal incidentaloma has a low pre-test probability of being an adrenocortical cancer or a phaeochromocytoma, but although both these tumours are potentially lethal they can be cured, or at least the patient's survival can be greatly improved by timely adrenalectomy (27-30).

In this report, our case was examined for dyspeptic symptoms unrelated to the adrenal gland and the ultrasonography determined a mass lesion in the adrenal compartment. As the clinical findings were nonspecific and originated from dyspeptic complaints, there were no findings of excessive hormonal secretions. Hormonal, biochemical and urine analysis were within normal limits. Dexamethasone suppression test with suppressed cortisol levels in this case indicated the phsiologic hypothalamopituitar-adrenal axes. As blood pressure, serum potassium and plasma renin activity were within normal limits, our patient was not thought to have excessive aldosterone. Urinary catecholamines were within normal limits. These findings confirmed that the mass in the adrenal region was non-functional and arised from extra-adrenal origin due to the outcomes of the clinical, biochemical, hormonal, and urine catecholamine tests extraadrenal lesion. For the radiological differential diagnosis, the images of sonography, computed tomography and MR defined that the lesion was related to accessory spleen. It was seen that there was no risk of malignancy or need for surgery.

The incidental discovery of adrenal masses raises the problem of distinguishing frequent benign masses from infrequent malignant ones (10-14, 19-21). Adrenal masses may be misdiagnosed as adjacent structures which may mimic adrenal masses. In this respect, accessory spleens are one of the differentials diagnosis to be considered. They are relatively common and are seen approximately in 2.5 – 30 % of the population (30-31). In a study where seven patients presented as adrenal incidentaloma, six were found to be accessory spleen and one was gastrointestinal stromal tumor which was confirmed by CT and contrast enhanced sonography (32). On CT images, accessory spleens are typically well-marginated, homogeneously enhancing round masses that are smaller than 2 cm (33) and there may not be a parenchymal bridge between the spleen and the accessory spleens like in our case.

Imaging modalities which will differentiate adrenal masses (adrenal adenomas, myelipomas, haemangiomas, ganglioneuromas, adrenal carcinomas, angiosarcomas, leiomyosarcomas, adrenal lymphomas, phaeochromocytomas, neuroblastomas and adrenal metastasis) and other left upper quadrant structures are very important. Computed tomography scan (unenhanced followed by contrast-enhanced examinations) is the cornerstone of imaging of adrenal masses (31). Attenuation values of <10 Housefield units on an unenhanced CT are practically diagnostic for adenomas (31).

When lesions cannot be characterised adequately with CT, MR imaging (with T1-and T2 weighted sequences and chemical shift and fat supression refinements) must be done (34-37). Functional nuclear medicine imaging is useful for adrenal masses that are not adequately characterised by CT and MRI. Scintigraphy with I-131 6-iodomethyl norcholesterol can differentiate adrenal cortical adenomas from carcinomas (38-40).

In the literature, several articles including case reports with accessory spleen were determined. In a patient who had previously undergone splenectomy, a biochemically inactive 5cm adrenal incidental mass was detected. In this case report, the authors suggest that radionuclide imaging with technetium sulphur colloid may provide information that would confirm the presence of accessory normal tissue and would therefore support observation rather than surgical resection (39, 40).

In other case reports, a soft tissue mass close to the upper pole of the left kidney was noted in a patient referred for intravenous pyelography. A left adrenal tumor was suspected. Computed tomography revealed an accessory spleen and the diagnosis was confirmed by abdominal angiography (41). In another report, a suspected left adrenal mass mimicking a carcinoma has been detected by US. A laparoscopic adrenalectomy has been performed. Following examination of the surgical specimen revealed that the resected adrenal gland contained no tumorous lesion. A further investigation of the intraperitoneal space has been revealed an accessory spleen (16, 17).

As in the present case report, when a mass in the adrenal region as an incidentaloma is determined, clinical, laboratorial and radiological examinations are sufficient to make a differential diagnosis. In conclusion, imaging findings are of the most important in making an accurate diagnosis in the difficult area of evaluating the masses in the adrenal localizations. The diagnosis of the accessory spleen mimicking a left-sided adrenal tumour as a differentiation for retroperitoneal and intraabdominal masses should be considered clinical, laboratorial and radiological, carefully.

Declaration of Interest

There is no conflict of interest that could be perceived as prejudicing the impartiality of the research reported.

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