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RENAL HEMANGIOMA: REPORT OF TWO CASES

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Renal hemangioma is considered a relatively rare disease. Virchow[1] described the first case from an autopsy specimen in 1867 and through 1967 more than 150 cases have been recorded in the world[2].

In Japan, the first case was found at an autopsy by Fukuda[3] in 1911 and Senzaki et al.[4] collected 34 cases from the literature up to February 1973. Recently we examined 2 patients having unusual signs and symptoms associated with the pathological demonstration of renal hemangioma. Some pathological and clinical observations are given about 37 cases in the Japanese literature including our 2 cases.

Pathological studies indicate that renal hemangioma may occur either in the renal parenchyma or pelvis. The parenchymal tumor is found in various locations, most frequently in the medulla.

Analysis of clinical studies shows that renal hemangioma does not always occur in patients under 40 years of age, in whom malignancy is uncommon. Preoperative diagnosis is still difficult, but the correct diagnosis can be made preoperatively using various clinical examinations.

CASE REPORTS

Case 1., T. T., a 26-year-old man, was admitted to our clinic with urinary retention due to bladder tamponade and with right flank pain on January 5, 1972. Past history revealed painless gross hematuria followed by bladder tamponade in September 1970, again January, November and December 1971. Family history was negative. On admission, his face was pale and the palpebral conjunctiva was slightly anemic. Physical examination revealed mild tenderness in the right upper quadrant and lower abdomen. Blood clots were seen at the external urethral orifice.

Immediate vesical lavage was carried out and a large quantity of coagulated blood was washed out. Cystoscopic examination demonstrated efflux of blood from the right ureteral orifice while clear urine from the left. Blood clots were found in the bladder, but the mucous membrane appeared normal.

Laboratory data were within normal limits except for a mild anemia with a red blood cell count of 392×10^4/cmm, a hemoglobin value of 77.5 per cent (Sahli) and a hematocrit value of 35.5 per cent.

Painless gross hematuria was seen for 2 days following admission after which the urine became clear. IVP demonstrated a filling defect of the right renal pelvis while the left kidney was normal (Fig. 1).

Right selective renal angiography showed a pooling shadow, 1.8 cm in the lower pole (Fig. 2). A radiographic deagnosis of right renal tumor was made and on January 17 a right nephrectomy was performed. The extirpated kidney measured 12 by 7 by 5.5 cm with a smooth, pale outer surface and demonstrated several small hematomas on the anterior surface. The capsule was easily stripped. Upon sectioning, there were bleeding foci at the tip of the pyramids and a hematoma could be seen at the

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lower cortico-medullary region (Fig. 3). The specimen demonstrated no evidence of malignancy despite the suspicious radiographic appearance. Histologic examination established an angioma of the kidney. Microscopically there were irregular sinuses lined by endothelium filled with red blood cells (Fig. 4).

Postoperative course was uneventful and the patient was discharged in good health 14 days after operation.

Case 2. T. F., a 67-year-old housekeeper was refered on February 16, 1970 for urological consultation because of a painless mass in the right upper quadrant. She had noticed the mass three months earlier, but hematuria or other urological symptoms had not been noticed. Past history and family history were nagative. Physical examination revealed the upper limit of liver dullness to be at the fourth intercostal space in the midclavicular line. The abdomen was flat and soft, but there was a smooth, firm and ill-defined mass which moved with respiration. The correlation between the mass and liver was not clear. The left kidney and spleen were not palpable. Laboratory data were normal

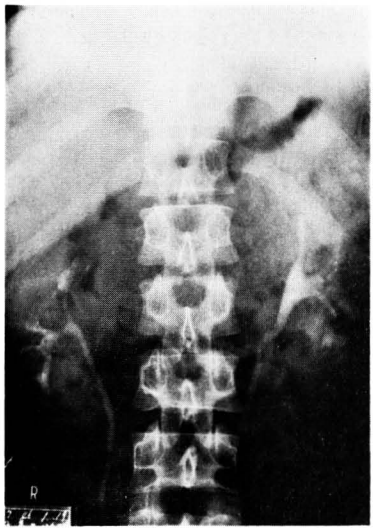

Fig. 1. Case 1, IVP is normal.

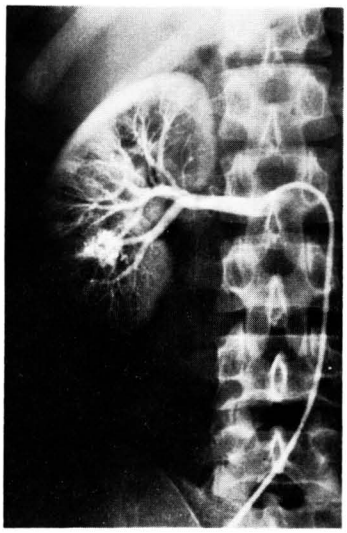

Fig. 2. Case 1, Selvective RAG of the right kidney: pooling shadow is found in lower middle lobe.

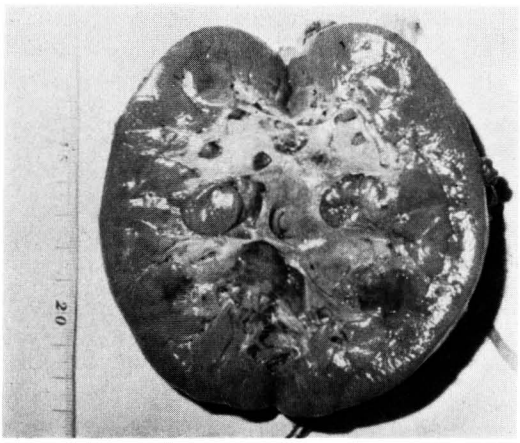

Fig. 3. Case 1, Section of specimen shows bleeding region, compatible with RAG findings.

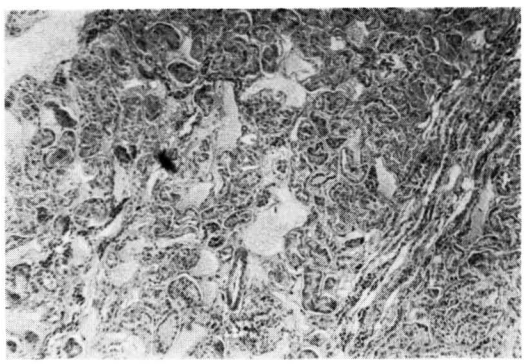

Fig. 4. Case 1, Histological findings show interstitial bleeding and nest of capillaries.

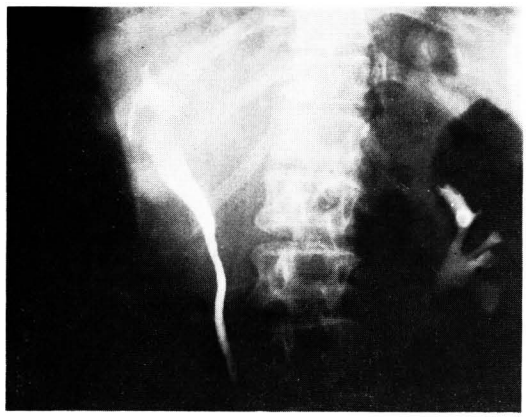

Fig. 5. Case 2, IVP shows deformity and filling defect of renal calyces and upward deviation of the right kidney.

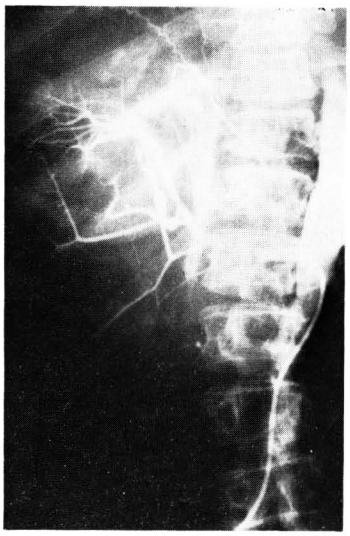

Fig. 6. Case 2, Selective RAG of the right kidney shows narrowing and stretched intrarenal arteries.

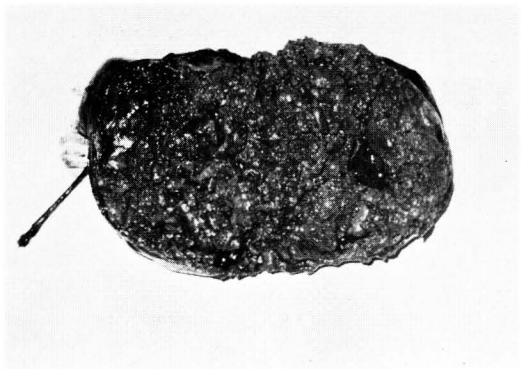

Fig. 7. Case 2, Section of specimen shows bleeding and abnormally grown vessels like sponge, no normal parenchyma.

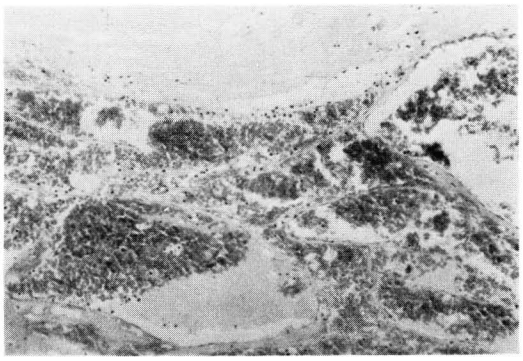

Fig. 8. Case 2, Histological findings show interstitial bleeding and dilated vessels.

except for a mild anemia with a red blood count of 376×10^4/cmm, a hemoglobin value of 62 per cent (Sahli) and a hematocrit value of 29.5 per cent. The urinary sediment showed ten to twenty red cells per high-power field and cystoscopic examination revealed normal bladder. IVP demonstrated poorly difined calyces of the right kidney, but the pelvis and the ureter were within normal limits; the left kidney and ureter showed a normal appearance (Fig. 5). Selective right renal angiography revealed narrowing of the renal artery and the intrarenal arteries poorly filled (Fig. 6). With the clinical diagnosis of the right renal tumor, on February 23, a right nephrectomy was performed. The specimen measured 15 by 15 by 15 cm and weighed 1750 g. The tumor was cystic and well encapsulated and the gross cut specimen showed the tumorous tissues had replaced the lower two-thirds of the kidney and was cavernous in appearance, filled with coagulated blood and necrotic material. There was a well-defined border between the tumor and the normal renal tissue of the upper pole (Fig. 7).

Microscopic examination showed large and small cavernous lumens, lined as a rule, by a single layer of endothelium (Fig.

Table 1. Cases of renal hemangioma in Japan from 1911 to 1973.

	AUTHOR	AGE	SEX	SIDE	C.C.	PYELOGRAM	RAG	CLIN. DIAG.
1	Fukuda (1911)	45	M	Lt.				nephritis
2	Fukuda (1911)	28	M					contracted kidney
3	Ohono (1923)	27 (19)	M	Rt.	hematuria	np		spont. renal bleeding
4	Kuroda (1949)	41 (39)	F	Lt.	hematuria	np		spont. renal bleeding
5	Abe (1952)	61 (61)	F	Lt.	hematuria	filling defect of upper calices		tumor of renal pelvis
6	Ohogoshi (1954)	27 (15)	M	Rt.	hematuria flank pain	np		spont. renal bleeding
7	Tanaka et al. (1955)	41 (41)	M	Lt.				spont. renal bleeding
8	Tuchiya et al. (1957)	48 (33)	F	Lt.	hematuria	unclear of upper calices		spont. renal bleeding
9	Doi et al. (1957)	20 (20)	F	Lt.	hematuria flank pain	deformity of renal pelvis		renal tumor
10	Nihira (1958)	34 (32)	F	Lt.	hematuria	np		spont. renal bleeding
11	Baba et al. (1961)	31 (21)	F	Rt.	hematuria	np	np	spont. renal bleeding
12	Tajiri et al. (1962)	53 (43)	F	Rt.	hematuria			spont. renal bleeding
13	Yamaguchi (1962)	30 (30)	F	Lt.	hematuria flank pain	filling defect of calices		benign renal tumor
14	Kosaka et al. (1964)	62 (52)	M	Rt.	hematuria	deformity of calices		tumor of renal pelvis
15	Takeuchi et al. (1965)	44 (44)	M	Rt.	hematuria	filling defect of P-U junction	np	tumor of renal pelvis
16	Miura et al. (1966)	53 (53)	F	Rt.	hematuria	np	A-V fistula like shadow	renal hemangioma
17	Sai et al. (1966)	18 (6)	F	Rt.	hematuria	filling defect of renal pelvis		tumor of renal pelvis
18	Minami et al. (1966)	23 (18)	F	Rt.	hematuria	delayed excretion	dilation pooling	renal hemangioma
19	Minami et al. (1967)	38 (28)	F	Lt.	hematuria flank pain ischuria	elongation of pelvis & calices	puddling	renal hemangioma
20	Minami et al. (1967)	64 (59)	M	Rt.	hematuria flank pain	deformity of middle calices	tortuous dilation pooling puddling	renal hemangioma
21	Minami et al. (1967)	27 (21)	F	Rt.	hematuria flank pain ischuria	np	tortuous dilation puddling	renal hemangioma

22	Takeda (1967)	63 (63)	F	Rt.	hematuria flank pain	deformity of lower calices		renal tumor
23	Ohogoshi et al. (1967)	58 (52)	F	Rt.	hematuria lower abdominal pain	filling defect of upper calices		tumor of renal pelvis
24	Mizumoto et al. (1967)	14 (13)	F	Lt.	hematuria	np	np	spont. renal bleeding
25	Harada et al. (1967)	26 (26)	M	Lt.	hematuria tamponade	deformity of lower calices	np	renal hemangioma
26	Takashima (1967)	54 (34)	F	Rt.	hematuria	filling defect of lower calices		renal tumor
27	Tokuhara et al. (1969)	34 (34)	M	Lt.	hematuria	no excretion of lower calices	np	renal tumor
28	Gotoho et al. (1970)	34 (20)	F	Lt.	hematuria	filling defect of lower calices	narrowing displacement	renal tumor
29	Senzaki et al. (1971)	66 (66)	F	Rt.	hematuria	filling defect of lower calyx		tumor of renal pelvis
30	Senzaki et al. (1971)	20 (20)	M	Lt.	hematuria	filling defect of renal pelvis	np	tumor of renal pelvis
31	our case (1971)	67 (67)	F	Rt.	tumor of hypochondrium	poor excretion	narrowing	renal tumor
32	Nakamura et al. (1972)	46 (46)	F	Rt.	hematuria	filling defect of renal pelvis	tumor vessels	renal tumor
33	Murayama et al.	58 (58)	M	Lt.	hematuria	np	curving circling avascularity	solitary cyst renal tumor
34	Fujita et al. (1972)	60 (60)	F	Lt.	hematuria	poor excretion of renal pelvis	np	tumor of renal pelvis
35	Senzaki et al. (1972)	64 (64)	F	Rt.	hematuria	filling defect of renal pelvis	np	tumor of renal pelvis
36	Itoh et al. (1972)	52 (52)	F	Rt.	hematuria	filling defect of lower calices	np	tumor of renal pelvis
37	our case (1973)	26 (25)	M	Rt.	hematuria tamponade	filling defect of middle calyx	pooling	renal tumor

C.C. : Chief complaint
RAG: Renal arteriography
( ) : age at onset of hematuria

8). Some of the larger cavernous lumens were obliterated by erythrocytes and blood pigments. In the central areas of the tumor, hyalinized connective tissue and blood clots were seen. Convalescence was uneventful and the patient remains well and asymptomatic postoperatively.

DISCUSSION

1) Frequency:

Hemangioma of the kidney is considered an exceedingly rare benign tumor. The first case was found at an autopsy by Virchow[1] in 1867. Riley and Swann[33] found no case of renal hemangioma in 13,219 autopsies in Boston City Hospital in 1939. Bell[34] observed only one case in 30,000 autopsies. Through 1967 more than 150 cases of this disease have been reported[2].

In Japan, Fukuda[3] described the first case from an autopsy in 1911, and Ohono[5] reported the first clinical case in 1923. Recently we collected 36 authentic cases after a review of the Japanese literature (Table 1).

2) Classification:

According to White and Braunstein[35], Anderson et al.[36] and Ferguson et al.[37], renal hemangioma is considered a true neoplasm rather than a dilatation of blood vessels. White and Braunstein[35] classified renal vascular tumors, excluding telangiectasis and varix, as follows.

(1) Benign : Capillary hemangioma, plexiform hemangioma and cavernous hemangioma.

(2) Malignant : Hemangiosarcoma, hemangioblastoma

And they considered renal hemangioma as one type of renal vascular tumor. As a rule, microscopically renal hemangioma takes the form of irregularly arranged thin-walled vessels of variable size lined by endothelium. The walls contain fibroblasts, angioblasts and newly formed capillaries[38]. However it is difficult to define by histological examination whether the renal hemangioma is a simple dilatation of normal vessels or a true vascular neoplasm. On the other hand, Dorman and Fowler[39] stated the differences between cases reported as hemangioma and teleangiectasia appear to be one of degree rather than kind. According to the classification of White and Braunstein[35], our first case is classified as a capillary hemangioma and the second case as a cavernous variant.

3) Location of lesion and size

Hemangioma occurs in every part of the kidney : cortex, medulla, pelvis, calyces, pyramids and papilla. Weyrauch and Berger[40] analysed the incidence of location in 76 cases as 48.7 percent in the mucosa and submucosal areas, 42.1 percent in the medullary portion of the kidney, and only 9.2 percent in the cortical area. In addition, Raff and Podolsky[41] reviewed 68 cases prior to 1947 and found 11 cases (16 percent) with renal pelvic lesions. In Japan, 5.8 percent are found in the medullary portion, 16.7 percent in submucosal area, 11.1 percent in both cortex and medulla, 8.3 percent in the pelvis and 5.6 percent in the cortical area (Table 2).

Table 2. Incidence of location of renal hemangioma in Japan.

	CASES (%)
RENAL PARENCHYMA (medullar, cortical and cortico-medullar portion)	28 (75.7)
SUBMUCOSAL PORTION AND RENAL PELVIS	9 (24.3)
TOTAL	37 (100.0)

Lesions have varied in size from pinhead and microscopic to measurements of 12 by 5 by 5 cm[2]. Lazarus and Marks[42] stated that the majority of tumors measure 1 to 2 cm in diameter. In our second case, the removed kidney was 15 by 15 by 15 cm in size and the tumor occupied over two-third of the kidney. This appears to be the largest in authentic cases reported in Japan.

Dorman and Fowler[39] commented that renal hemangioma usually occur singly, although multiple tumors in the same kidney have been reported[43].

4) Age, sex and side of lesion

Patients with clinically apparent renal hemangioma ranged between 4 days old

Table 3. Age and sex distribution and incidence of affected side.

SEX	AGE						TOTAL
	0 - 20	21 - 30	31 - 40	41 - 50	51 - 60	61 - 70	
MALE	1 (3)	5 (2)	1 (1)	3 (2)	1 (3)	2 (0)	13 (11)
FEMALE	3 (5)	3 (4)	4 (4)	3 (2)	6 (4)	5 (4)	24 (23)
TOTAL	4 (8)	8 (6)	5 (5)	6 (4)	7 (7)	7 (4)	37 (34)

( ) : age at onset of hematuria, except for unknown 3 cases

SEX	SIDE			TOTAL
	Rt.	Lt.	UNKNOWN	
MALE	6	6	1	13
FEMALE	14	10	0	24
TOTAL	20	16	1	37

and 12 years old with a peak between 30 and 40[44,45]. In 85 percent of the cases studied by Riley and Swann[33], the onset of hematuria occurred in patients before the age of 40. This is in contrast to the incidence of malignant tumors of the kidney which occur predominantly in patients older than 40. However, Wallach et al.[46] stated that in 87 cases from the literature, only 45 cases (52 per cent) occurred before the age of 40 and there was no correlation between age and occurrence of this disease. Minami et al.[25] reviewed 16 clinical cases from the literature in Japan and commented that only 9 cases (56 per cent) were found before age 40, but in 73 per cent of the cases the onset of hematuria occurred in patients under 40 years of age. Whereas in our study, in 33 clinical cases from the literature, only 19 cases (57.5 per cent) experienced the onset of hematuria before the age of 40 (Table 3). In general, cases are nearly equally distributed between male and female[39]. Anderson et al.[36] stated that more cases had been reported in men rather than in women. In Japan, however, women have been involved slightly more than men (Table 3). Peterson et al.[2] stated that the right and left kidney are equally involved. The same finding is demonstrated in our study as shown in Table 3.

5) Symptoms

Hematuria is the first and frequently the only symptom of renal hemangioma and varies from microscopic to large amounts of blood. The onset is usually sudden and without any obvious causes in patient in seemingly good health. Hematuria is usually intermittent. And the interval between these recurring attacks may be a few days to many years. The longest duration of hematuria was seen in the case reported by Rotlino and Mohan[47], and Anderson et al.[36] reported a case with long interval of hematuria. In our study, the longest duration of hematuria was 15 years.

Table 4 shows the duration of hematuria of 33 authentic clinical cases of Japan and in 9 of these hematuria persisted for over 10 years. In our second case, the chief

Table 4. Duration of hematuria in renal hemangioma.

DURATION (YEARS)	CASES (%)
WITHIN 1	16 (47.1)
1 ---- 3	4 (11.7)
4 ---- 6	4 (11.7)
7 ---- 9	1 (3.0)
OVER 10	9 (26.5)
TOTAL	34 (100.0)

(except for unknown 3 cases)

complaint was presence of a mass in the right hypochondrium and the patient had no obvious hematuria.

Pain in the upper quadrant not infrequently accompanies the hematuria and is caused by partial obstruction of the renal pelvis by clot. Ureteral colic is produced by the passage of blood clots and may be more common in older people[38]. There is often a temporary disturbance in urination e. g. frequency and urgency due to intravesical blood clots. In our first case, hematuria followed by dysuria was the main symptom.

6) Diagnosis

It is usually very difficult to confirm the diagnosis of renal hemangioma by clinical examination before surgery. Almost all cases recorded to the date have been diagnosed postoperatively by the pathologist after nephrectomy for probable malignancy. According to Dorman and Fowler[39], Dean and McCarthy reported an unusual case associated with multiple hemangioma of the skin, in which the preoperative diagnosis of renal hemangioma was confirmed by the pathologist. Later Butt and Perry[48] recorded a case diagnosed preoperatively. In the Japanese literature, Minami et al.[21] and Miura et al.[18] reported the cases in which a preoperative diagnosis had been made. In the authenticated cases, spontaneous renal bleeding was found preoperatively in 25 percent (Table 5). According to Minami et al.[21], Dukes performed nephrectomy on 9 patients under the diagnosis of spontaneous renal bleeding. Of these 9 patients, renal papillary angiomas were found in 3 patients, and he suggested that renal hemangioma was the most common cause of so-called spontaneous renal bleeding. In 1941, Webb-Johnson and Turner-Warwick[49] reported the difficulty of demonstrating angioma of the renal papillae in cases of essential hematuria. However, every case of spontaneous renal bleeding, if studied intensively enough, will reveal some underlying pathological condition sufficient to explain the bleeding. In order to make the correct diagnosis of this disease, several special examinations have been carried out, eg pyelography, angiography renoscintigraphy and Papanicolaou staining of urine sediment. Among them, renal angiography may play an important role. Anderson and Rasmussen[50] reported a case of hemangioma of the renal pelvis, in which the diagnosis was made preoperatively by selective renal angiography and they stated that sharp demarcation, densely arranged vessels of uniform calibre, regular outline and the marked intensity of the medium in the capillary part of the growth, were recognized as benign features that did not occur in malignancy.

Table 5. Preoperative diagnosis of renal hemangioma in Japan.

PREOPERATIVE DIAGNOSIS	CASES (%)
TUMOR OF RENAL PELVIS	10 (27.0)
SPONTANEOUS RENAL BLEEDING	9 (24.3)
RENAL TUMOR	9 (24.3)
RENAL HEMANGIOMA	6 (16.3)
BENIGN RENAL TUMOR	1 (2.7)
UNKNOWN	2 (5.4)
TOTAL	37 (100.0)

7) Treatment

Nephrectomy is the treatment of choice if the lesion is unilateral and the opposite kidney has good function. This results from the difficulty in making the correct preoperative diagnosis as well as the control of hematuria often associated with other benign diseases, and in differentiating renal hemangioma from disease requiring nephrectomy such as carcinoma. Heminephrectomy and papillectomy may be indicated to some cases, provided that the focus in small, well localized and not malignant. Because these two operations, however, are not always successful, in almost all cases nephrectomy was performed. In our study of Japanese literature, all cases received nephrectomy because of above reasons. Because radiotherapy is valuable in th etreatment of benign vascular tumor, radiotherapy should be tried on renal hemangioma for the purpose of

conservation of renal parenchyma[46].

SUMMARY

Two cases of renal hemangioma with unusual symptoms are described. In the first case the chief complaint was recurrent dysuria due to blood tamponade, and in the second case it was the right upper quadrant mass without hematuria.

A review of the cases recorded in the literature of Japan has been carried out. Renal hemangioma does not necessarily occur in patients older than 40 years in whom malignancy are often seen.

Correct preoperative diagnosis of this disease may be difficult in spite of various special clinical examinations. In one-forth of the authentic cases, the preoperative diagnosis has been spontaneous renal bleeding. Some authors consider that renal hemangioma is the most common cause of this sign.

As a rule, nephrectomy is the treatment of choice due to the difficulty in making an accurate preoperative diagnosis.

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腎血管腫の2例

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腎血管腫はまれな疾患の1つとされており，本邦報告例のほとんどが腎摘除後に確認されている．われわれは最近，膀胱タンポナーデをくり返し腎腫瘍の疑いで腎摘除術を施行したが組織学的に腎血管腫と診断された1症例ならびに腹部腫瘤を主訴とした1症例を経験したので報告する．

症例1：26歳，男性．主訴は膀胱タンポナーデによる尿閉．1971年1月，11月，12月に膀胱タンポナーデを起こして救急入院しているが詳細不明のまま経過し，1972年1月5日ふたたびタンポナーデを訴えて当科に入院した．入院時理学的所見では瞼結膜蒼白，右側腹部に軽度の圧痛あり，膀胱洗浄にて多量の血塊を排泄した．同時におこなった膀胱鏡所見では粘膜正常，右尿管口より出血を認めた．IVP所見は正常で選択的右腎動脈撮影では右腎中下部に径約1.8 cm大の濃染部を認めpooling像を思わせる陰影が得られた．しかし腎静脈がとくに早く出現する像は認められなかった．以上の所見より右腎腫瘍の疑いで腎摘除術を施行した．摘出腎は175 g，12×7×5.5 cmで表面平滑であったが，ところどころに小血腫がみられ，割面では下腎部の乳頭と皮髄境界部に出血巣を認めた．組織学的所見は異常血管のnestと尿細管の入り乱れた像でrenal hemangiomaのcapillary typeと診断された．

症例2：67歳，家婦．主訴は右上腹部の無痛性腫瘤で約3カ月前より同腫瘤を触れていたが血尿など泌尿器系疾患に由来する症状は認めていない．理学的所見では肝濁音界が右鎖骨中線上第4肋間腔まで上昇しており右上腹部の腫瘤は表面平滑，弾性硬，境界不鮮明で呼吸性移動を認めた．膀胱鏡所見は正常，IVPでは左腎は正常であったが，右腎杯の描出不良で腎盂，尿管は正常であった．選択的右腎動脈造影では腎動脈は細く，腎内動脈は枯枝状であった．右腎腫瘍の疑いで腎摘除術を施行，摘出腎は15×15×15 cm，1750 gで割面では腎の約2/3が海綿状の腫瘍組織で占められており，血塊および壊死組織が充満していた．組織学的所見は1層の内皮細胞で覆われた大小さまざまなlumenが海綿状にみられ，renal hemangiomaのcavernous typeと診断された．

以上腎血管腫の2例を報告するとともに本邦報告例37例について若干の考察を加え，また欧米での報告例とも比較検討を試みた．

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