Teratomas in Children: Review of Cases in the 0-12 year age Group seen at Harari Hospital over Ten Year Period, 1960-1969

BY

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INTRODUCTION

Much literature has been published on teratomas in childhood from many centres throughout the world (Willis (1951), Engel *et al.* (1968), Kemp (1967), Hickey and Layton *et al.* (1954), Abell *et al.* (1965), Bale (1950), O'Leary and Halfort (1947), Arnheim, Cohen *et al.* (1950), Silberman and Mendelson (1960), and Gwinn *et al.* (1955)), and yet little from this country. Ross and Bell (1971) published a report of two cases of ovarian teratomas causing gliomatosis peritonei; one was a six year old girl. In an earlier paper Ross (1967), made brief mention of teratomas in the Mashonaland African and remarked that they were predominantly sited in the sacral area and ovary. No other literature has come to the writer's notice on teratomas in the Mashonaland African.

AIM

This paper is a review of the teratomas seen at Harari Central Hospital and Central Histological Laboratory in the ten year period from 1960 to 1969 in the 0-12 years age group. All the cases were Africans from the area of Rhodesia lying to the north of a line drawn through Que Que, Umvuma, Gutu and Chipinga — Ross (1967), conducted his survey in this area.

SCOPE OF REVIEW

Human histological specimens from the above mentioned area are sent to the Central Histological Laboratory at Harari Hospital; patients from this area are also referred to Harari Central Hospital for specialist treatment and services. Most cases under review were either patients who had been referred to this hospital or born here; the rest were laparatomy specimens sent in for histological diagnosis.

TABLE I.

Gives the Sex, Age, Site, Presentation and Special Features.

Number	*Sex*	*Age*	*Site*	*Presentation*	*Special Features*
2093/60	F	3 days	Sacrococcygeal	Mass behind anus since birth	Benign
1022/61	F	12 years	Sacrococcygeal	Discharging sacral sinus for one year	Benign
1896/61	F	1 month	Right neck	Swelling right neck since birth	Benign
2674/62	F	2½ years	Retroperitoneal	Swelling left abdomen for six months. No pain.	Benign
267/63	F	6 days	Sacrococcygeal	Mass since birth	Benign
2425/63	F	5 years	Neck	Midline swelling since birth, getting bigger	Benign
2520/63	F	10 months	Sacrococcygeal	Mass present at birth, getting bigger	Malignant
2299/63	F	12 years	Ovary	Lower abdominal swelling and pain	Benign
3385/63	F	3 days	Sacrococcygeal	Mass since birth	Benign
818/64	F	3 days	Sacrococcygeal	Mass since birth	Benign
1035/64	F	10 months	Left neck	Swelling left neck since birth, getting bigger	Benign
3146/64	M	2 years	Sacrococcygeal	Mass since birth	Benign
3275/64	F	7 years	Sacrococcygeal	Mass since birth	Benign
204/65	F	7 days	Sacrococcygeal	Mass since birth	Benign
1734/65	F	3½ months	Sacrococcygeal	Mass since birth	Benign
1434/65	F	4 months	Sacrococcygeal	Mass since birth	Benign
2970/65	M	7 days	Sacrococcygeal	Mass since birth	Benign
1171/66	M	9 months	Sacrococcygeal	Mass since birth	Benign, sacral spina bifida also present
2309/66	M	1¾ years	Sacrococcygeal	Mass since birth	Benign
4442/66	F	11 years	Ovary	Recent lower abdominal pain and swelling	Benign
767/67	F	2 weeks	Sacrococcygeal	Mass since birth	Benign
3200/67	F	2 years	Sacrococcygeal	Swelling since birth	Ectopic anus. Malignant
3643/67	M	1 month	Sacrococcygeal	Mass since birth	Benign. Associated meningomyelocoele present
4080/68	F	4 months	Abdomen	Semi cystic cenral abdominal swelling since birth	Benign
1768/69	F	1 year	Sacrococcygeal	Mass since birth	Malignant
2440/69	M	12 years	Right ischial fossa	Very haemorchagic tumour in ano for one month	Malignant. Healed surgical scar over sacrum
4355/69	F	6 years	Ovary	Recent abdominal swelling with pain	Benign. T.B. Peritonitis queried
4754/69	M	2 years	Perineal	Perineal mass arising from base of bulb of urethra	Malignant
4833/69	F	8 days	Sacrococcygeal	Mass since birth	Benign

Method

All the histology reports on teratomas in the relevant age group and seen during the period in question were obtained from the Central Histological Laboratory; the original slides and paraffin block were also obtained. The relevant case notes were obtained from the Medical Records Office at Harari Hospital and from the National Archives.

The original slides, stained with haematoxylin and eosin were reexamined, not so much to rectify the initial diagnosis but to take full stock of the tissues present and also to look for possible malignant changes and features. Where possible the original paraffin blocks were examined using the multiple section method as recommended by Willis (1951); and where technically possible sections were taken at 90° to the original slides. All these were stained with haematoxylin and eosin; where necessary special stains were employed. Sections were examined under the ordinary light microscope. Particular search was made for malignancy, firstly because the clinical presentation suggested malignancy and secondly because of the relative rarity of malignant change in teratomas in children.

The case notes were consulted as to the history, presentation, site and follow up where possible.

Findings

Twenty-nine (29) teratomas were seen between 1960 and 1969. Twenty-four were benign and five malignant i.e. embryonal carcinomas. The benign neoplasms contained tissues from the three germinal layers, but in varying amounts. See Table I. The numbers used are those on the histology report. The case notes did not give the measurements of these tumours.

Discussion

The number of teratomas seen do not give an absolute reflection of the incidence of these neoplasms in the population and age group under question. Writers have often remarked on the reluctance of the local population to seek orthodox medical treatment, or if any at all, Ross (1967). Therefore not all such cases were seen.

Secondly since most cases less than a week old were delivered in medical institutions and referred to this hospital; those that were delivered at home by the traditional midwife may not have presented themselves for orthodox medical treatment. Resort to traditional forms of medicine and their subsequent and relative failure and unsatisfactory results; and the long distance to be travelled in the case of referred patients, may in part account for the relatively late age of presentation at Harari Hospital.

Much literature has been published on sacrococcygeal teratomas. Willis (1951), and Gross *et al.* (1951), have written extensively on the nature and possible actiology of these neoplasm. Jackman *et al.* (1951), and Gwinn *et al.* (1955) have written on the anatomy of the sacrococcygeal area.

The sacrococcygeal area is the commonest location for the majority of teratomas in children. Nearly all are present at birth and according to Conklin and Abell (1967), their incidence is 1/40 000 live births. Most papers published show a preponderance of these neoplasms in females Arnheim (1952). Conklin and Abell (1967), in their series of 32 cases found that 78 per cent. were females. Willis (1951) made the same observation. Hickey and Layton (1954), found that in their series of 40 cases 94 per cent. were females. In this series there were 19 sacrococcygeal teratomas; all were present at birth and 74 per cent. were females.

TABLE II.

Shows the Distribution According to Sex and Site.

Site	*Females*	*Males*
Sacrococcygeal	14	5
Ovary	3	0
Neck	3	0
Abdomen	1	0
Retroperitoneal	1	0
Perineum	0	2
Total	22	7

Tissues from the three germinal layers were fully represented. It is of interest to note that two teratomas were found in association with sacral spina bifida and another with a meningomyelocoele; both cases were males. This relationship is well documented. Hickey *et al.* (1954), in their series had three cases of sacrococcygeal teratomas who had sacral and lumbar spina bifida and meningomyelocoele. This association has also been noted by Gwinn *et al.* (1955), Gross *et al.* (1951), Riker and Potts (1942), Alexander and Stevenson (1946) and Jackman *et al.* (1951). Other series have emphasised the frequent association with twinning or family history of twinning and malformations. (Hickey *et al.* (1954)). Malignant sacrococcygeal teratomas are discussed later.

Ovarian teratomas (dermoid cysts) are usually present at birth but present later in life. According to Willis (1951), "the delay in discovery is readily understood, they are usually benign and slow growing and are so situated that they are unlikely to cause symptoms until they attain considerable size." Abell *et al.* (1965), and Groeber (1963), have published articles on ovarian tumours in children and have come to the conclusion that teratoma of the ovary in the pre-pubertal girl is relatively uncommon. In this series there were three cases of ovarian teratomas, which is about 11 per cent. of all teratomas seen.

One case, a six year old girl, presented with what was thought to be a tuberculous peritonitis with lower abdominal swelling. This turned

out to be gliomatosis peritonei; this is peritoneal disemmination and implantation of benign teratomatous tissues which is at times mistaken for malignant change Willis (1951). This particular case was reported by Ross and Bell (1971). Another case had marked bilharzial infection. The third was unremarkable. No malignant features were seen in these three teratomas.

Teratomas of the neck are relatively uncommon; only three were seen in this series, and were in females. Two were on either side of the neck and the third was in the midline. Histology shows no thyroid tissue in these neoplasms. Bale (1950) published a review article on these tumours.

In 44 of the 56 cases he stated that part or all of the thyroid gland was replaced by the tumour and that it was no pressure effect but actual absence of the gland with the tumour occupying its space. He admits that identification of thyroid tissue in these is unusual, he quotes six in 56, about 11 per cent. Silberman and Mendelson (1960), quotes a figure as high as 40 per cent. Bale (1960), stressed the close developmental relationship of the thyroid and these tumours; he observed that some of these tumours got their blood supply from the thyroid. Silberman and Mendelson (1960), found that 16 per cent. of cases of teratomas of the neck were still births.

Various authors have recommended that the usage of the term "teratomas of the thyroid region" and not "teratomas of the thyroid gland" because not all teratomas of the neck are associated with the thyroid gland nor do they all replace the thyroid gland nor do they all contain thyroid tissue.

One case of retroperitoneal teratoma was seen in this series. O'Leary and Halport (1947), stated that benign neoplasms with the structure of a teratoma are exceedingly rare in this site. Arnheim (1952), states that such neoplasms are the third commonest cause for retroperitoneal swelling in infancy, the more common being neuroblastomas and nephroblastomas. From the articles by Arnheim (1952), O'Leary and Halport (1947), Cohen *et al.* (1950), Longino and Martin (1958), and Engels *et al.* (1968), it is evident that these neoplasms are relatively rare, and commonest in females. Arnheim (1951), reviewed all the reported cases in literature and found that 26 were females, 16 males and two were not stated; 19 were in the right side, 11 on the left and 14 bilateral. The one case in this present series was a girl and the teratoma was on the left side.

Teratomas of the abdominal viscera (apart from gonads) are very rare; the abdomen is the least common site for such tumours, Willis (1951).

Teratomas of the kidneys have been described by various writers; Langley (1950), McCurdy (1943) and McDonald (1968). In this series one case of an abdominal teratoma was found in a four year old girl. The neoplasm was attached to the lesser curvature of the stomach and the liver. It was benign.

There were five teratomas which contained embryonal carcinomatous elements. See Table III. They were classified as embryonal carcinomas; no attempt was made to split them up into groups, as advocated by Dixon and Moore (1952).

TABLE III.

Shows the Distribution According to Sex and Site of the Benign Teratomas.

Site	*Females*	*Males*
Sacrococcygeal	11	5
Ovary	3	0
Neck	3	0
Abdomen	1	0
Reptroperitoneal	1	0
Total	19	5

TABLE IV.

Shows the Distribution According to Sex and Site of the Embryonal Carcinomas.

Site	*Females*	*Males*
Sacrococcygeal	3	0
Perineal	0	2
Total	3	2

Much literature has been written on embryonal carcinomas and carcinomatous change in teratomas in childhood; Lisco (1942), Ravich

et al. (1966), Holtz and Abell (1963). From what has been done and written on this subject, it is evident that the chances of malignant change in a benign teratoma are increased if there is delay in treatment. "Tumour removed after five months of age either because of their late appearance or because of delay in the institution of therapy are most apt to embryonal carcinomatous elements, at least two-thirds of those treated in this age period are biologically malignant" (Nelson). It has also been observed that in a few instances surgical meddling or incomplete removal of an apparantly benign neoplasm seems to stimulate it into a hopeless malignancy. Gross *et al.* (1951). In this series the age range was from ten months to 12 years with the mean age of three and a half years, which falls within the age period already referred to.

Two of the three sacrococcygeal embryonal carcinomas developed in teratomas that were present at birth. The 12 year old boy who presented with an embryonal carcinoma of the perineum, was found to have an old healed surgical scar on the sacrum which seemed to suggest that a sacral teratoma had been removed at some time but the parents denied previous operation or sacral mass. This was, however, put down in the case notes as either malignant change following surgical intervention or a recurrence.

The two year old girl who presented with a sacrococcygeal embryonal carcinoma also had a ectopic anus. She was successfully treated then but came back nine months later with recurrences of the malignancy. Nothing could be done for her.

It is likely that the malignancy in the two year old boy that arose from the base of the bulb of the urethma could have been from testis, but the case notes make no mention of the state of the testes.

Summary

Twenty-nine cases of teratomas were seen in this series; 24 were benign and five were embryonal carcinomas. Of the benign neoplasms 16 were sacroccoccygeal, three ovarian, three in the neck, one abdominal and one retroperitoneal. These findings are in agreement with the literature reviewed, in that the sacrococcygeal area is the commonest location for teratomas in infancy and childhood and that the other sites are not so common. Three embryonal carcinomas were seen in the sacrococcygeal area and two in the perineum.

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