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Sacrococcygeal teratoma in children (about 26 cases)

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ABSTRACT:
Introduction: Sacro-coccygeal teratomas are the most frequent tumors of the infant, the diagnosis used to be made at the time of delivery what created major obstetrical problems. Thanks to the progress of medical imaging, the diagnosis is now made prenatally.
Material and method: We report a series of 26 cases documented in the department of pediatric surgery at the children's hospital of Rabat over a period of 22 years from 1998 to 2020.
Results: 77% of our patients presented with a mass. No associated malformation was noted. An extension evaluation was positive in 3 patients.
Surgical treatment consisted of total resection of the tumor with coccygectomy. 13 patients required chemotherapy.
Discussion: Sacrococcygealteratomas are germ cell tumors, the most frequent extra gonadal location.
These tumors develop pre-sacral and retro-rectal on the midline. In the neonate the development is most often exopelvic, while in the older child the development is endopelvic, which explains the delay in diagnosis and the revelation by signs of compression of the pelvic organs.
The prognosis of TSC diagnosed before 2 months is generally good.
Conclusion: Sacro coccygeal teratomas are frequent germ cell tumors. We report the experience of our department in the management of these tumors whose treatment is essentially surgical and, in some cases, may require chemotherapy.
Keys words: Sacro-coccygeal teratomas, germ cell, tumor, coccygectomie.

I. INTRODUCTION:

Sacro coccygeal teratomas (SCT) are the most prevalent neonatal tumors. The discovery of this disease was usually made during childbirth, but now and thanks to the progress of medical imagery, it is possible to diagnose it prenatally and to provide a more efficient approach to the treatment of the disease.

we report 26 cases of SCT collected in the department of pediatric surgery at the University Hospital of Rabat.

II. MATERIAL AND METHODS:

We received 26 SCT files from the registry of the pediatric surgery department of the University Hospital of Rabat . The data were processed by SPSS software

III. RESULTS:

The mean age was 17.87 months with the extremes of age from 6 days to 72 months
Sex ratio M/F= 0.44
Rate 1.24/year
88% of patients (n=23) were delivered vaginally while 12% of patients (n=3) where delivered by cesarean section
1 case was diagnosed antenatally, at birth in 12 cases, before 1 year in 3 cases and after 1 year in 10 cases.

The warning signs were diverse:
Sacroccygeal mass 77%, urinary manifestation 15.4%,digestive manifestation 27%,Neurological manifestation 23%,behavioral disorder 7.7%.
On clinical evaluation, the tumour mass size varied from 2.5 cm to 23 cm with a mean of 8 cm.
An X-ray of the pelvis was performed in 13 patients, finding an opacity of fluid tonality with intratumoral calcification in 5 cases
An abdominopelvic ultrasound showed hypoechogenic heterogeneous masses with double or triple component of tissue echostructure in 25 patients
in 19 patients the evaluation was completed by a CT scan revealing a sacrococcygeal mass and 5 patients underwent an MRI, one of them antenatally
The extension test identified metastasis in 3 patients

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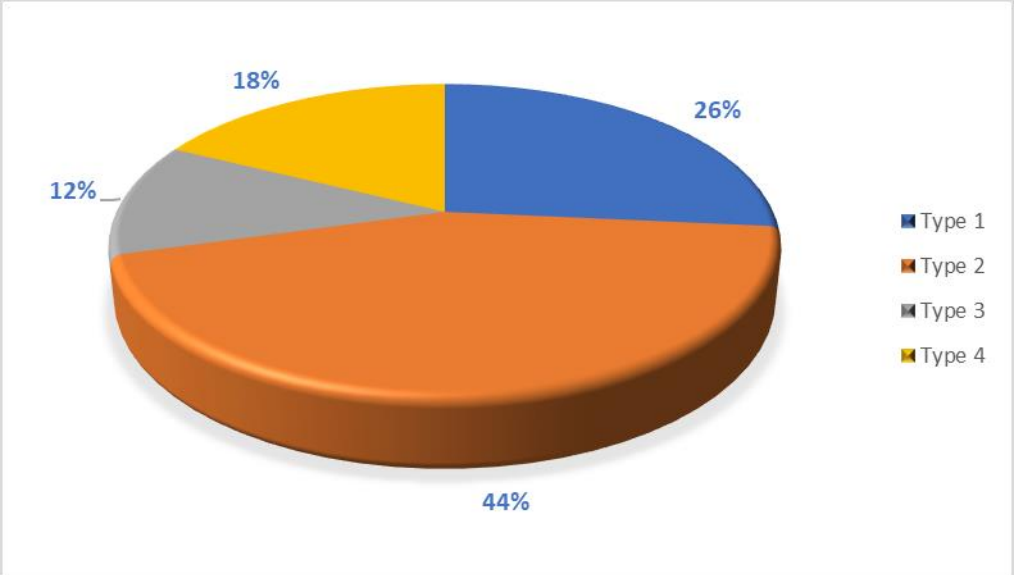
IV. CLASSIFICATION:

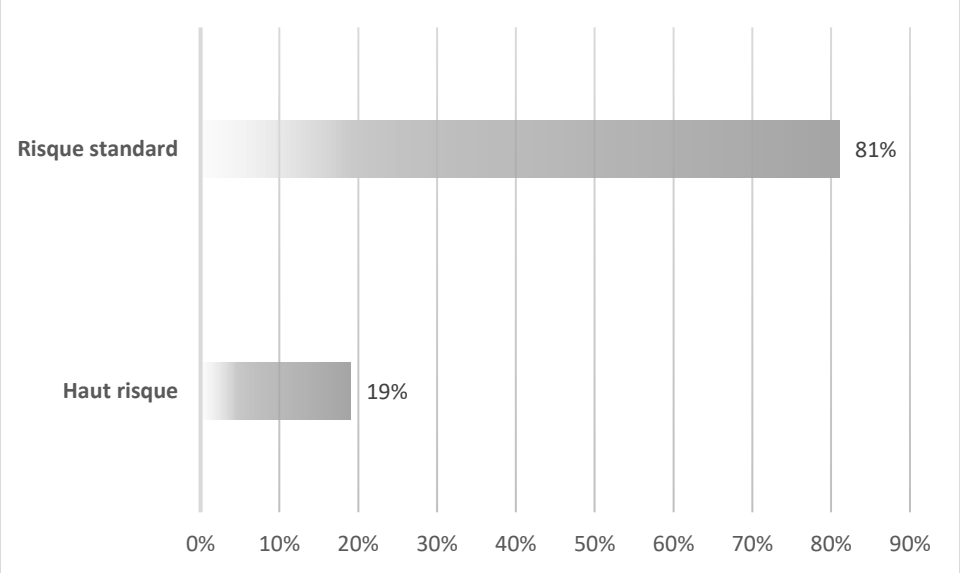


Classification according to ALTMAN

Biomarkers:

Alpha feto protein positive		**Alpha feto protein negative**
< 15000 IU/ml	>15000 IU/ml	8 cases
14 cases	4cases	

Risk-based classification:
This classification is based on the level of Alpha feto protein and/or the presence of metastasis



Risk-based classification
Treatment:
In our series, 13 patients benefited from surgery only whereas 13 patients had surgery combined with chemotherapy.
Surgery:

Total tumor resection with coccygectomy by perineal approach in 21 patients and by abdominoperineal approach in 5 patients

Chemotherapy:

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4 cases pre and post operative; 1 post operative, and 8 pre operative.

Histology:

Mature teratoma		15 cases
Immature teratoma		5 cases
Malignant teratoma	Yolk Tumor	5 cases
	Embryonic carcinoma	1 case

Repartition according to the histological type

Evolution:

Immediate course: Wound superinfection (1 case) rectal fistula (1 case)

Medium- and long-term evolution:Encoparesis and neurological bladder (1 case);Recurrence 11 cases; death 2 cases; good evolution 15 cases; 6 patients lost to follow-up.

V. DISCUSSION:

Sacrococcygeal tumors are rare tumors with an overall incidence of 1/35-40000 living births (1).The sacrococcygeal localization is the first place of neonatal teratomas and the first place of neonatal solid tumors. (2-3) .A clear female predominance with a sex ratio of 4G/1B (36). Studies have shown that there are familial forms with autosomal dominant transmission (4). There would also be a considerable predominance in twins. (5)

The average age of discovery is 22 months, but the forms diagnosed at birth are the most frequent. (6)

In our study,1 case was diagnosed antenatally, at birth in 12 cases, before 1 year in 3 cases and after 1 year in 10 cases.

There may be several associated midline conditions such as vertebral column, urogenital, digestive, cardiac, cleft lip or cleft palate malformations (7)

Despite its characteristic appearance, the sacrococcygeal teratoma can be confused with other masses that develop in the sacrococcygeal region, such as myelomenigocele, horsetail lipoma, cystic lymphangioma, rhabdomyosarcoma, pelvic neuroblastoma, ependymoma, chondroma.... (8) from which the anatomopathological study takes its place.

It has been suggested that 25% of sacroccygealteratomas are mixed with a benign and malignant component (9). The prevalence of malignancy is proportional to age (10).

In terms of management, sacroccygealteratoma is treated as soon as it is diagnosed, in the ante-natal period by methods of tumour reduction by total exeresis in utero after hysterotomy (11), or endoscopically by laser phacocoagulation (12), echo-guided radiofrequency (13), diathermic loop thermo-coagulation (14), and embolisation of the vascular pedicle (15).

The moment of delivery of a newborn with a sacroccygealteratoma is a critical moment and must be well planned and agreed upon by the obstetrician, intensive care, and pediatric surgery teams. (16)

The basis of treatment for sacrococcygealteratoma is surgical resection from the first days of life to minimize the development of malignant components; total coccygectomy is systematic to avoid recurrence. (17)

Tumor resection should not obscure the need for reconstruction of the perineal muscles to maintain normal anal continence, and skin reconstruction to ensure a satisfactory aesthetic appearance of the gluteal region. (17)

The surgery can be followed by adjuvant treatment for malignant teratomas (18)

Sacroccygeal teratomas have the highest risk of recurrence of all germ cell killers, ranging from 7 to 35% (19). In literature, immature teratomas have a higher recurrence rate than mature teratomas. The histological type of recurrent teratomas is not always similar to that of the primary tumor (20).

VI. CONCLUSION:

Sacroccygeal teratomas are congenital tumors depending on the caudal pole of the embryo, composed of 3 different embryonic cell layers. The incidence of malignancy increases markedly with age, making early diagnosis essential. Treatment can be carried out in utero, thanks to the advent of ante-natal Doppler, which helps to clearly identify the vascularization of the tumor.

Treatment at birth is surgical, with total exeresis combined with coccygectomy.

Patient follow-up is reinforced during the first year and attentive over the following 3 years.

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