Multiple Presacral Teratomas in an 18-year-old Girl: A Case Report

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Although the sacrococcygeal area is the most common site for a teratoma in infants, it is a rare site for a teratoma in older patients. Most of the teratomas found in this area in adults are single mass, but in a few cases, multiple masses have been reported. The author reports on the case of an 18-year-old female patient with 3 presacral teratomas. The tumors were surgically removed via a transabdominal approach and were pathologically diagnosed as mature cystic teratomas. This case report indicates that an adult presacral teratoma can appear as multiple tumors, although it is very unusual.

Keywords: Adult presacral teratoma; Multiple

INTRODUCTION
Teratomas are composed of various cell types, which represent more than one germ layer. Sacrococcygeal teratoma is the most common solid neoplasm in neonates, with an estimated prevalence of 1 in 35,000-40,000 births. They can be diagnosed prenatally by fetal ultrasound; 50-70% of cases are found during the first few days of life, with less than 10% being diagnosed beyond the age of 2 years [1]. Case reports on adult teratoma in this anatomic site are very rare. Complete surgical resection is necessary to alleviate symptoms and to rule out malignancy because malignant transformation has sometimes been reported [2]. Almost all reported cases of adult teratomas have been found as a single tumor, so multiple masses are extremely unusual. We report a case of adult teratoma found as multiple tumors in the presacral area.

CASE REPORT
An 18-year-old female was transferred to our outpatient depart-
filled cysts. Sebaceous material was also aspirated from the cyst in order to obtain a better operative view. The solid portion contained mainly fatty tissue and calcifications (Fig. 3). Histopathological evaluation revealed a vast spectrum of tissue types characteristic of a teratoma. All three germ layers appeared in all 3 tumors: fragments of squamous epithelium, hair, sebaceous glands, and neural glia (ectodermal derivation); adipose tissue and bone (mesodermal derivation); and respiratory epithelium (endodermal derivation). No malignant or immature cells were found, and a diagnosis of mature cystic teratoma was made. The patient recovered without showing any postoperative complication.

Fig. 1. (A) CT scan of the pelvis demonstrating two masses in the presacral space. Note calcifications (arrow) within fat tissue in the left cyst. (B) Coronal view showing three discrete masses.

Fig. 2. The specimen shows three well-encapsulated masses. Since the cystic contents were partially aspirated during dissection, the size of the mass seen in the specimen is smaller compared to the original one.

Fig. 3. (A) Hair, (B) sebaceous material, (C) fatty tissue, and (D) calcifications are seen within the cystic mass.
DISCUSSION

Teratomas contain tissues that are foreign to their anatomic site and have not resulted from metaplasia. They are derived from embryonic pluripotent cells and may have various degrees of matura-
tion, according to which they are classified as mature, immature, and malignant [3]. They may be inherently malignant or have the potential for malignant degeneration. Among the pediatric pop-
ulation, there is a tendency toward malignant transformation of sa-
crococcygeal teratomas with increasing age. The incidence of ma-
lignancy is 7-10% when diagnosis is made prior to the age of 2
months, compared with 50-67% when the diagnosis is established 
after that age [4]. However, benign tumors predominate in case reports involving adult populations [5, 6]. The possibility of ma-
lignancy should be kept in mind when planning the treatment for adult sacrococcygeal or presacral teratomas because cases of ma-
lignant transformation have sometimes been reported [2, 5, 7].

With regard to radiologic evaluation, the presence of irregular calcifications has been reported in 75% of benign tumors [8]. Cal-
cifications are also found in 25% of malignant teratomas, so they cannot be considered to be an indicator of benignity [9]. Malign-
nancy is suspected in large tumors with necrosis, poor definition of adjacent soft tissue planes, and sacral infiltration and is certain when locoregional lymph node and distant metastases are noted [8, 9]. Based on this information, the tumor in the present case was considered to have a benign nature in preoperative evaluation, although imaging features alone do not provide definite differentiation between benign and malignant teratomas. In this case, benignity was confirmed using an intraoperative frozen biopsy.

Most teratomas contain both solid and cystic areas, although com-
pletely solid teratomas do occur. Teratomas are usually well-encap-
sulated single masses, and independently encapsulated cysts can rarely be found within one large tumor [10]. However, the appear-
ance of an adult sacrococcygeal or presacral teratoma as multiple masses is extremely rare, and according to a thorough literature review, only one report has shown a case of presacral teratoma ex-
hibiting three cystic masses [11]. Various theories for the origin of teratomas include parthenogenetic development of germ cells within the gonads or in extragonadal sites; nonparthenogenetic origin from “wandering” germ cells left behind during migration of embryonic germ cells from the yolk sac to the gonad; and an origin from other totipotent embryonic cells [3]. Pluripotent cells are normally present in the gonads and may also be found in abnormal sequestered midline embryonic rests. Accordingly, terato-
mas are found in the mediastinum, the retroperitoneal space, the sacrococcygeal zone and intracranial locations, as well as the go-

nads. If the pathogenesis of sacrococcygeal or presacral teratomas that are aberrantly sequestered pluripotent cells is considered, the presence of multiple tumors might be possible, as in the reported case, which is very unusual.

Symptoms of presacral teratomas are often subtle and nonse-
cific; however, completely asymptomatic patients, like the present case, are rare. Most frequently reported symptoms include lower back and pelvic pain, constipation, urinary retention and lower extremity paresthesias or weakness [7, 11]. Symptoms mainly re-
sult from the mass effect and rarely from infiltration in the case of malignancy. In fact, these symptoms can be elicited by any type of presacral mass. Differential diagnoses of presacral masses vary with their natures. In adults, presacral simple cystic lesions may corre-
spond to anterior meningoceles or rectal duplication cysts. In the appropriate clinical context, they may also represent abnormal col-
lections, such as seromas or urinomas. In the presence of a multi-
loculated cystic lesion, a tail gut cyst must be considered. Denser and more complex lesions may represent chronic retrorectal ab-
scesses, pilonidal or dermoid cysts, soft tissue, bone tumors and metastatic tumors. Whatever the nature of the lesion is, a presacral or sacrococcygeal teratoma should be included in the differential diagnosis [10].

CT and MRI are the most significant methods to characterize the mass and to evaluate the intrapelvic extension and relationship to other structures. Both studies are complementary; however, CT is the most sensitive study for demonstrating calcification and ossi-
fication in sacrococcygeal teratomas, as well as for determining the integrity of the adjacent cortical bone. MRI allows a better topographic evaluation of the tumor owing to its direct multipla-
nar characteristics and provides higher resolution in soft tissues. In this case, MRI was not performed because the boundary of the mass was clear on the CT scan. However, for a thorough preoper-
ative study in complicated cases, MRI could be necessary.

Surgical excision is the treatment of choice for presacral terato-
mas, provided that the tumor can be completely removed. A pos-
terior approach through a sacral incision, a transabdominal ap-
proach and a combined approach have been reported, depending on the tumor’s size and location. The transabdominal approach is preferred for high lesions without evidence of sacral involvement. This approach has the advantage of providing excellent exposure of important pelvic structures. Recently, laparoscopic excision has also been attempted by some particular surgeons [12, 13].

In conclusion, a presacral teratoma should be considered in the differential diagnosis of a pelvic mass in adults. An adult teratoma at this site can appear as a multiple tumor, although it is extremely unusual.

CONFLICT OF INTEREST

No potential conflict of interest relevant to this article was reported.

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