ISSN 0008-3194 (p)/ISSN 1715-6181 (e)/2015/21–25/$2.00/©JCCA 2016

Benign sacrococcygeal teratoma incidentally found on routine scoliosis radiographs in a 12-year-old female: a case report

Kelly M. Cunningham, MD, MA, BSc[1]
Govind B. Chavhan, MD, DABR[2]
Kelly E. Ainsworth, DC, MD, FRCP(C)[3]

Objective: *To describe the imaging characteristics of sacrococcygeal teratomas and to review appropriate diagnostic evaluation and management.*

Clinical Features: *12-year-old otherwise healthy, asymptomatic female with an incidental pelvic mass found on routine scoliosis radiographs.*

Intervention and Outcome: *The pelvic mass was further evaluated by MRI and CT scan. Management consisted of successful surgical resection with no post-operative complications. Pathology confirmed a mature, benign, sacrococcygeal teratoma.*

Conclusions: *Chiropractors manage patients with scoliosis, which may include radiographic surveillance. Familiarity with the radiographic features of masses such as sacrococcygeal teratomas is important for prompt diagnosis and management.*

(JCCA. 2016;60(1):21-25)

KEY WORDS: sacrococcygeal teratoma, pediatric

Objectif : *Décrire les caractéristiques d'imagerie des tératomes sacrococcygiens et examiner l'évaluation et la prise en charge du diagnostic approprié.*

Caractéristiques cliniques : *Jeune fille asymptomatique de 12 ans en bonne santé avec une masse pelvienne découverte fortuitement après des radiographies de routine pour la scoliose.*

Intervention et résultats : *La masse pelvienne a davantage été évaluée à partir d'images d'IRM et de TDM. La solution était la résection chirurgicale réussie sans complications postopératoires. La pathologie a confirmé un tératome sacrococcygien mature bénin.*

Conclusions : *Les chiropraticiens prennent en charge les patients atteints de scoliose, ce qui peut inclure la surveillance radiographique. La familiarité avec les caractéristiques radiographiques des masses, comme les tératomes sacrococcygiens, est importante pour leur diagnostic et la prise en charge rapide.*

(JCCA. 2016;60(1):21-25)

MOTS-CLÉS : tératome sacrococcygien, pédiatrique

1 Department of Diagnostic Imaging, McMaster University, Hamilton, ON, Canada
2 Department of Diagnostic Imaging, The Hospital for Sick Children and University of Toronto, ON, Canada
3 Department of Diagnostic Imaging, McMaster University Medical Centre and McMaster University, 1200 Main Street West, Hamilton, ON, L8N 3Z5, Canada, email: ainswork@hhsc.ca

Corresponding author:
Kelly E. Ainsworth
Department of Diagnostic Imaging, McMaster University Medical Centre and McMaster University
1200 Main Street West, Hamilton, ON, L8N 3Z5, Canada
Email: ainswork@hhsc.ca

Written consent to participate in this case study was provided.

Introduction

Mature cystic teratomas are defined as neoplasms comprised of well-differentiated derivations of two or more germ cell layers (ectoderm, mesoderm and endoderm). They are well known for their diverse anatomic locations and can occur anywhere from the brain to the gonads, however they occur most commonly along the midline (mid-axial).[1] In children, as in our case presentation, these tumors are often extra-ovarian, in contrast to an ovarian predilection in adults.[2] Mature teratomas are the commonest germ cell tumor, and are often incidental findings.[5] Incidental discovery on radiographic evaluation for routine chiropractic investigations would therefore not be considered an isolated or rare event, although their incidence appears to be underreported in the literature. The purpose of this report is to describe the imaging characteristics of an incidental sacrococcygeal teratoma found on routine surveillance scoliosis radiographs and to review appropriate diagnostic evaluations and management.

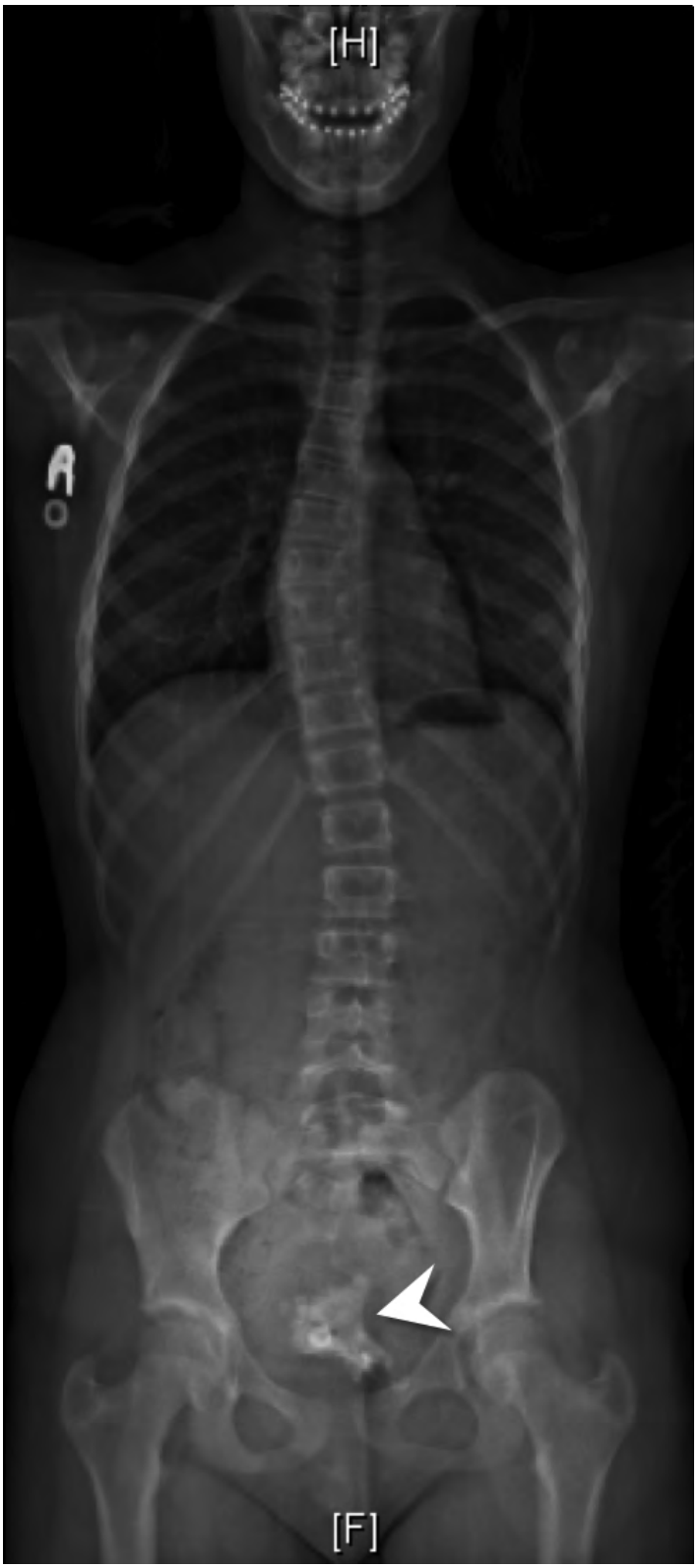
Methods & Results

A literature search was performed using PubMed with the search terms, "teratoma" AND "case report" AND "radiograph" as well as "teratoma OR dermoid" AND "incidental" AND "case report". No similar case reports were found. Images were obtained from the institutional picturing archiving and communication system (PACS). The patient's electronic medical record was reviewed for details on patient demographics, clinical history, operative report and pathology results.

Case Presentation

Routine spine radiographs were obtained on a 12-year-old, otherwise healthy, asymptomatic girl for follow-up of scoliosis. Incidental note was made of a large, well-defined, heterogeneous mass within the pelvis containing calcified and likely ossified densities (Figures 1A, 1B). Radiographs from one-year prior did not include the pelvis so chronicity could not be determined. Given the unclear etiology of this density and suspicion for an underlying mass, further evaluation was performed.

An unenhanced MR of the pelvis was obtained, using axial and coronal T1 spin echo, and axial and sagittal T2 fat saturation fast spin echo sequences (Figures 2A, 2B). MRI demonstrated a large (11.0 x 12.1 x 14.6 cm), complex, pre-sacral mass, which contained fat, calcium, and



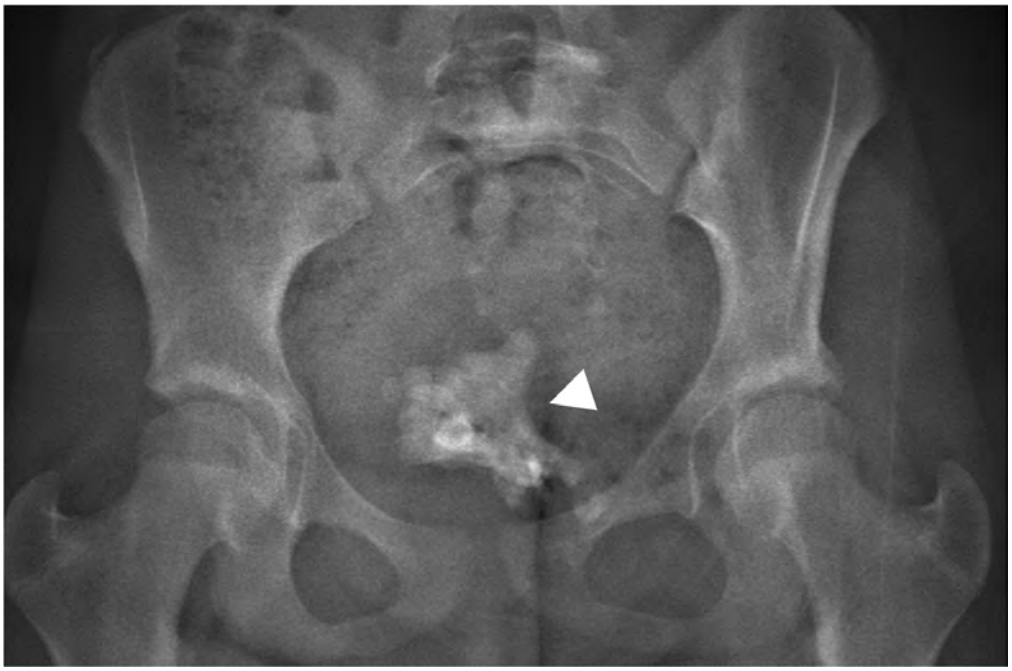


Figure 1: *(page 22) A. Posteroanterior full spine radiograph shows a calcified, and likely ossified, amorphous pelvis mass (arrowhead). (right) B. Cropped, magnified view of pelvic mass.*

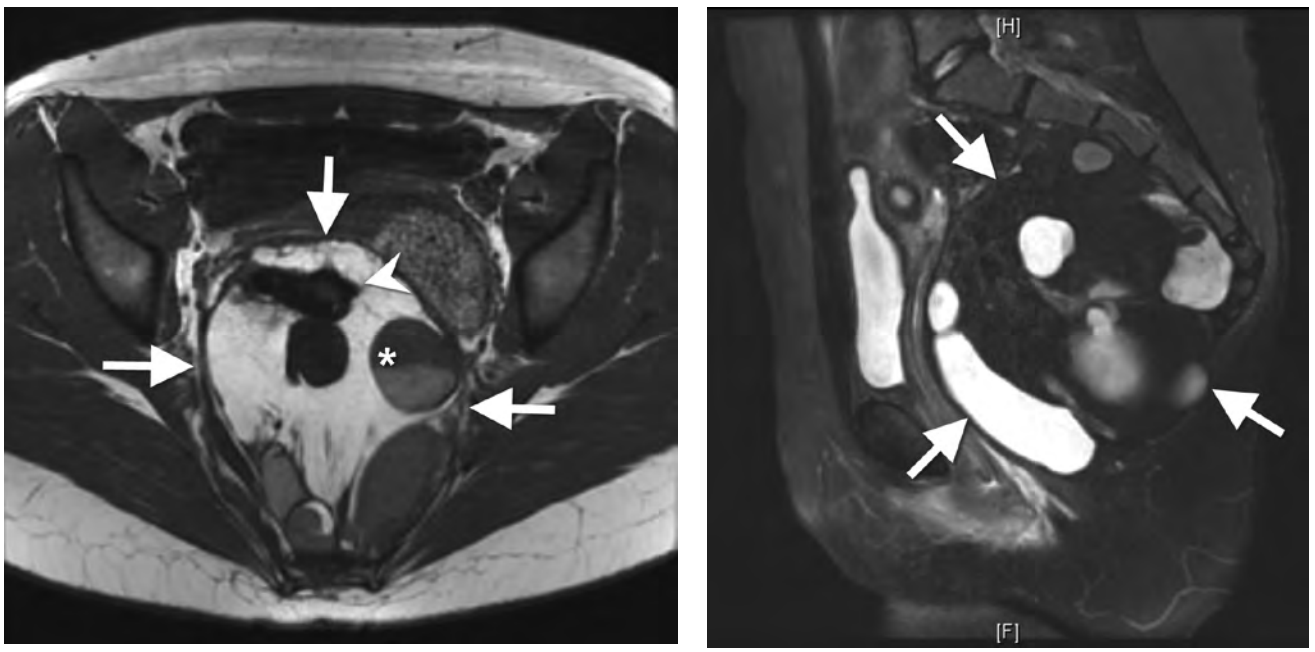


Figure 2:
A. Axial T1 Spin Echo, B. Sagittal T2 Fat Saturated (FS) Fast Spin Echo MR images of the pelvis show a large heterogeneous presacral mass (arrows). The mass shows a component with fluid-fluid level (asterisk) and calcified component (arrowhead).

cystic components (both clear and hemorrhagic), one of which contained a fluid-fluid level. The mass extended from the level of S2 to the anal canal, displacing the rectum anterolaterally, and the vagina and bladder anteriorly. The uterus was displaced anteriorly and to the right. The left ovary was clearly visualized in the left iliac fossa and a small right ovary was suspected adjacent to the right iliac vessels. There was no direct extension to the spine, and no signal abnormalities were seen within the bony sacrum. These findings were felt to be most consistent with a non-ovarian, mature pre-sacral teratoma. Gynecologic consultation was recommended for clinical evaluation and management.

Further evaluation with CT was performed to better characterize the calcifications and for pre-operative planning. CT confirmed the presence of a large heterogeneous mass with macroscopic fat (asterisk), calcific foci and a central, ossified bony component (arrowhead) (Figure 3). A reasonable adjunct in work-up could include pelvic ultrasound, although this was not performed in this particular case.

The patient was taken to surgery and the mass was excised. There was no involvement of the bony sacrum or soft tissue extension beyond the pelvic cavity. The mass contained a central bony core which had to be divided and removed in piecemeal. Both ovaries were visualized and deemed to be preserved and uninvolved.

Pathologic assessment of fresh gross surgical specimen revealed a large fragmented mass, composed primarily of fibroadipose tissue and bone. Hemorrhagic components and hair were visualized within the specimen. The mass had a maximal dimension of 20cm, with the bony fragment measuring 7.5cm in length. The final diagnosis was a mature sacrococcygeal teratoma with degenerative changes.

Discussion

Incidentally discovered teratomas in various clinical settings have been described[3], however no cases have been described related to incidental discovery on routine scoliosis radiographs. Given the gravity of potential complications, such as bowel obstruction, ovarian torsion, hemorrhage and rupture, and consequently, peritonitis, it is important for the chiropractic community to familiarize themselves with the typical radiographic appearance and epidemiology of mature pelvic teratomas.

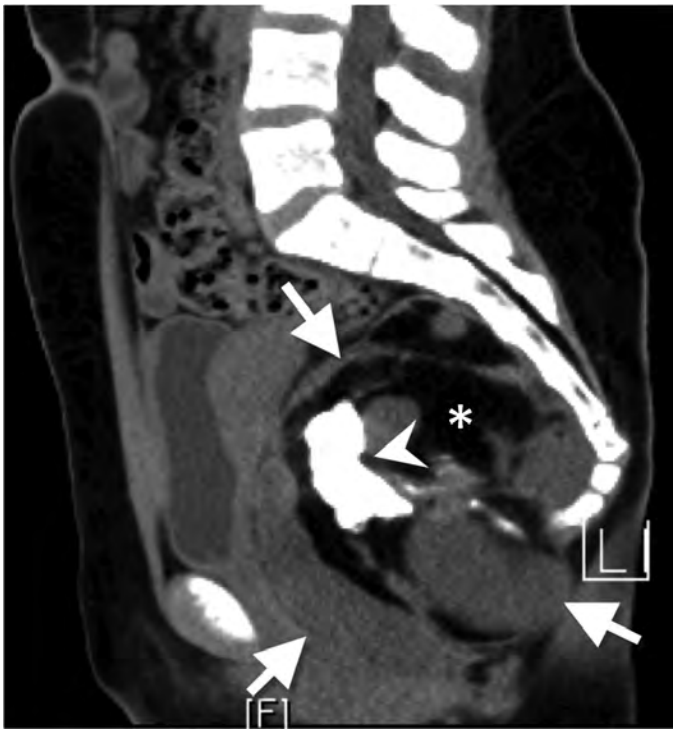


Figure 3:
Sagittal unenhanced CT image of the pelvis shows the presacral mass (arrows) containing fat (asterisk), calcification (arrowhead) and soft tissue. The calcified components show hyperdense attenuation on CT image.

In a large study of childhood teratomas conducted at an exclusively pediatric medical centre, Bale et al. describe and classify 107 teratomas over a 40-year period.[2] There was a significant female predilection (63%) which is consistent with the general literature. Sacrococcygeal teratomas accounted for 48% of cases.[2] They further subdivided the sacrococcygeal masses into posterior (with no intra-pelvic component), pelvic and dumbbell (tumor straddles pelvic and posterior compartments). The majority (67%) were situated posteriorly. The diagnosis of the pelvic subtype is usually delayed (diagnosed after 2 years of age).[2,4] Using Bale's criteria our case would be further characterized as a pelvic (or pre-sacral) teratoma as there was no extra-pelvic extension. Teratomas can be benign (mature) or malignant. The malignant potential (roughly 20% of lesions) increases with the internalization of tumor such that the risk of malignancy is greater in the pelvic subtype.[2,4] Malignant tumors were almost

exclusively carcinomatous. Conversely, the incidence of malignancy fell with posterior/external location. Benign teratomas are predominantly cystic, and contain mature tissue including fat, calcification and a small volume of solid soft tissue.[4] The larger the volume of solid, immature elements increases the risk of malignant potential[4] and the size of immature teratomas was larger than that of mature teratomas[5]. Histologic maturity did not vary by age of the patient.[5]

Tapper and Lack further described 254 benign teratomas assessed at a pediatric medical centre over a 54-year period.[1] Similarly, most were detected in the newborn period, had a strong female predilection (84%) and the majority were sacrococcygeal in location (40%).

Teratomas are commonly found within the pelvis and typically contain a variety of tissues such as hair, calcium and fat. Their classic radiographic appearance is that of a pelvic mass containing tissue of fatty density, often with calcification (and/or ossification), though it can be quite variable.[7] CT often confirms the presence of cystic, fatty and bony components. MR will again demonstrate fat signal within the lesion and drop out on fat saturation sequences. Cystic components will frequently contain a fluid-fluid level, specifically fat-fluid, primarily from liquid sebum.[7] While the differential of a fat and calcium containing pelvic mass is quite limited, one could consider the possibility of malignant degeneration of a teratoma, or an atypical liposarcoma. However, given the classic imaging features in our case, the radiologic diagnosis was most consistent with that of a mature cystic teratoma.

The size of the mass in our case presentation (20 cm) may have been underestimated by imaging which was reported as 14.6 cm in maximal dimension. Unfortunately, the true size of the mass may have been difficult to confirm as it had to be removed in piecemeal during the surgery. The gross specimen size was larger than measurements reported in the literature[1], which describe an average diameter of 7.5 cm in mature teratomas of the sacrococcygeal region.

Our report describes a large, mature pelvic-subtype sacrococcygeal teratoma which was found incidentally in an asymptomatic 12-year-old girl. The epidemiologic features are considered classic, with patient age and tumor size at presentation both being greater than that of the general literature.

Significant risks associated with pelvic teratomas include ovarian torsion, spontaneous rupture, hemorrhage, bowel obstruction, infertility, pregnancy complications, and malignant transformation.[5] Although the malignant potential is low, given this, as well as the other serious potential complications, and to prevent local recurrence, definitive and complete surgical excision of the lesion using minimally invasive and fertility-preserving techniques, remains best practice at all ages.[1,5,6] For this reason, further evaluation with CT or MRI and referral to a gynecologist or general surgeon for definitive management is indicated once the diagnosis of teratoma is suspected.

Conclusion

Mature, cystic teratomas are the most common type of germ cell tumor. Location depends on age, occurring primarily in the sacrococcygeal region in children and in the ovary in adolescents and young women. While they are typically benign they do demonstrate a small malignant potential and can cause severe complications such as bowel obstruction, ovarian torsion and peritonitis. Identification and intervention is therefore important for optimal management. Given their location, young age at presentation, as well as their indolent and often asymptomatic course, this presents a unique opportunity for the chiropractic community to recognize these lesions during routine skeletal evaluations such as scoliosis radiographs.

References:

1. Tapper D, Lack EE. Teratomas in infancy and childhood. Ann Surg. 1983; 198: 398-410.
2. Bale PM, Painter DM, Cohen D. Teratomas in childhood. Pathology. 1975; 7: 209-218.
3. Hong JH, Lee JK, Song SH, Hwang JH, So KA, Shin BK, Lee NW, Lee KW. Unilateral ovarian dermoid cyst accompanied by an ipsilateral paratubal cyst in a girl with Proteus Syndrome discovered by laparoscopic surgery. J PediatrAdolesc Gynecol. 2010; 23: 107-110.
4. Kocaoglu M, Frush DP. Pediatric presacral masses. Radiographics. 2006; 26: 833-857.
5. Comerci JT, Licciardi F, Bergh PA, Gregori C, Breen JL. Mature cystic teratoma: a clinicopathologic evaluation of 517 cases and review of the literature. Obstet Gynecol. 1994; 84:22-28.
6. Oelschlager AEA, Sawin R. Teratomas and ovarian lesions in children. Surg Clin North Am. 2012; 92: 599-613.
7. Dodd GD, Budzik RF. Lipomatous tumors of the pelvis in women: spectrum of imaging findings. AJR. 1990; 155: 317-322.