

CASE REPORT

Complete spontaneous regression of a subpubic cartilaginous cyst : a case report

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Abstract : Subpubic cartilaginous cyst was recently reported as a rare degenerative mass on the pubic symphysis. We report here a 59-year-old woman who presented with a vulvar mass that showed complete spontaneous regression 48 months after the initial visit. Treatment was only wearing brace. This is the first report of complete spontaneous regression of a subpubic cartilaginous cyst. In the case of small subpubic cyst, observation and follow-up alone may be sufficient. *J. Med. Invest.* 63 : 319-322, August, 2016

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INTRODUCTION

Subpubic cartilaginous cyst was first reported in 1996 by Algucial-Garcia and Littman (1). It is a rare tumor-like mass that contains degenerative cartilaginous tissues arising from the pubic symphysis (1). A few case reports have described the cyst to be stable in size (2) (3) or to reduce only slightly in size over time (4), with no reports of cases where the tumor regressed. This report presents a first case of subpubic cartilaginous cyst that showed complete spontaneous regression.

CASE REPORT

A 59-year-old woman presented to her gynecologist with a 2-year history of a vulvar mass with dull pain. Ultrasonography revealed a tumor in the pubic symphysis and she was subsequently referred to our department for further examination.

Physical examination revealed an immobile vulvar mass. Plain radiography showed slight sclerotic changes around the pubic symphysis. Magnetic resonance imaging (MRI) revealed a 1.5×1.0×1.0 cm mass located anterior to the middle pubic symphysis (Figure 1). The lesion presented as a low-intensity area on T1-weighted images and as a homogeneously high-intensity area on T2-weighted images. The lesion also showed high-intensity attenuation on short T1 inversion recovery images. Gadolinium enhancement was not observed in the mass except peripheral lesion. Positron emission tomography/computed tomography showed no 18-fluorodeoxyglucose accumulation in the soft tissue mass on the anterior pubic symphysis, but showed bone erosion of the anterior pubis. These findings suggested the possibility of a subpubic cartilaginous cyst.

Since the patient had only dull pain, follow-up observation by MRI was decided. She had continuous dull pain and the tumor still remain at 18 months after her initial visit and she started wear a pelvic brace. Forty-eight months after her initial visit, repeat MRI showed the cyst had completely disappeared and the dull pain also

disappeared (Figure 2).

DISCUSSION

Subpubic cartilaginous cyst was first reported by the pathologists, Algucial-Garcia and Littman, in 2 patients who presented with an unusual type of subpubic cystic mass (1). They described a tumor formed from fibrocartilaginous tissue with extensive cystic degenerative changes. This benign cyst appeared to be related to, and perhaps originating from, the periarticular tissues of the pubic symphysis. Although a few cases have been reported since, there have been no other cases in which the tumor completely regressed, as in the present case.

We reviewed 12 cases of subpubic cartilaginous cyst reported in the literature to date (Table 1) (1-10). Almost all cases involved multiparous females in their 50's, 60's, and 70's who presented with vulvar masses and various symptoms. Four cases presented as painless vulvar mass (1), 3 as a painful mass or abdominal pain (2), 4 as urinary dysfunction (1, 9), and in one rare male case as pain in the basis of the penis and sexual dysfunction (4).

In 2004, Kim *et al.* described the MRI features of a subpubic cartilaginous cyst (5). The lesion had a broad margin of contact with the adjacent pubic symphysis. In later publications it was described as hypointense relative to muscle on T1-weighted sequences, heterogeneously hyperintense on T2-weighted sequences, and with a thin enhancing wall with no internal enhancement (2). Plain radiography showed a relatively wide pubic symphysis with sclerosis (7). These MRI and radiographic findings are relatively specific, meaning that diagnosis should be possible based on such typical image findings.

The differential diagnosis includes aggressive anigomyxoma, non-ovarian cyst, urethral diverticula, vulval carcinoma, and chondrosarcoma (3) (6). Aggressive anigomyxoma (11) is one of the benign tumor affecting the pelvis and perineum. The tumor has a well-defined margin and shows hypointense in T1-weighted images and high intensity in T2-weighted images. This tumor has a specific feature, a swirled appearance, and hypervascular unlike subpubic cartilaginous cysts. Non-ovarian cyst (12) such as Nabothian cysts, Bartholin's cyst, and Gartner's cyst, is not associated with the pubic symphysis. Urethral diverticula (13) are anatomically different from subpubic cartilaginous cyst. Cancer of the vulva (14), accounts for 3-5% of primary gynecologic malignancies,

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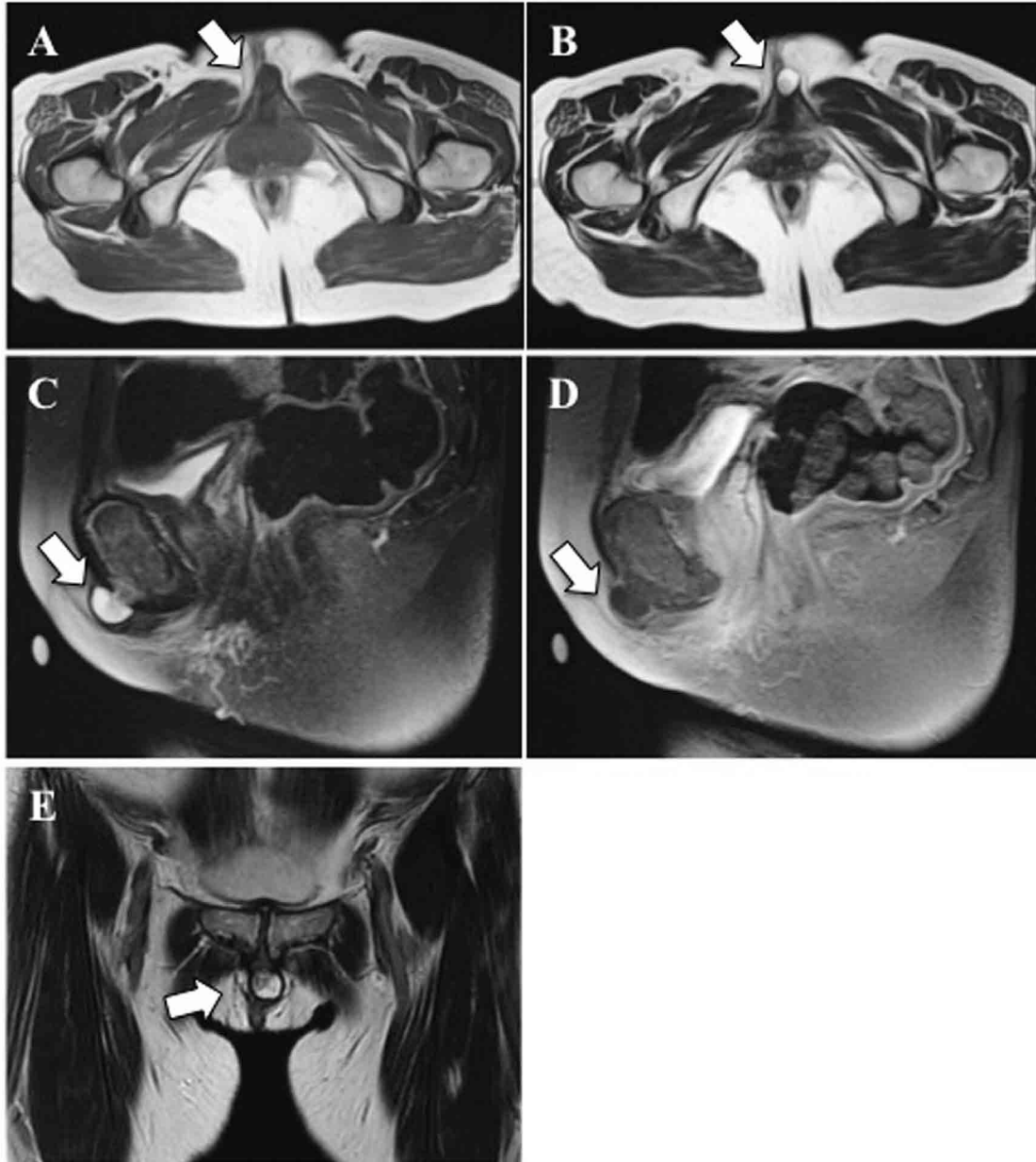


Figure 1 MRI images show a 1.5×1.0×1.0 cm mass anterior to the middle pubic symphysis (white arrow). A : Axial T1- WI shows low-intensity attenuation ; B : axial T2- WI and C : sagittal STIR images show high-intensity attenuation ; D : sagittal contrast-enhanced MRI showing no gadolinium enhancement of the tumor except for peripheral lesion ; E. coronal T2- WI. MRI, magnetic resonance imaging ; WI, weighted imaging ; STIR, short T1 inversion recovery

was isointense to muscle on T1-weighted images and showed intermediate-to-high signal intensity on T2-weighted scans. Fifty percent of cancer of the vulva showed enhancement. The location was different from subpubic cartilaginous cyst because they are not associated with the pubic symphysis. Chondrosarcomas are rare primary bone tumor. Large and multi-lobulated mass in pelvis can mimic chondrosarcoma (3).

Although needle biopsy (2) and open biopsy (3) were performed in 2 cases, biopsy may be limited to difficult cases only, such as those with a huge mass with intrapelvic and extrapelvic components that had a possibility of sarcoma (3). Performing biopsy from an anatomically difficult location may cause tumor contamination if it is sarcoma. In cases where biopsy is difficult, a metastable technetium-99

methylene diphosphonate whole-body bone scintigraphy would be helpful as it demonstrate mild radiotracer uptake within the symphysis pubis, but none in the parasymphyseal mass itself (3). These bone scintigraphy findings may indicate degenerative changes. Moreover, no 18-fluorodeoxyglucose accumulation in positron emission tomography/computed tomography indicate a little possibility of malignancy. The diagnosis of bone and soft tissue tumor are difficult, therefore, it is better to consult radiologist and orthopedic surgeon who major in musculoskeletal oncology.

Treatment for subpubic cyst is observation or resection. Almost all cases that presented with symptoms, such as urinary retention, underwent tumor resection ; observation was chosen in almost all asymptomatic cases. Cyst aspiration and instillation of local

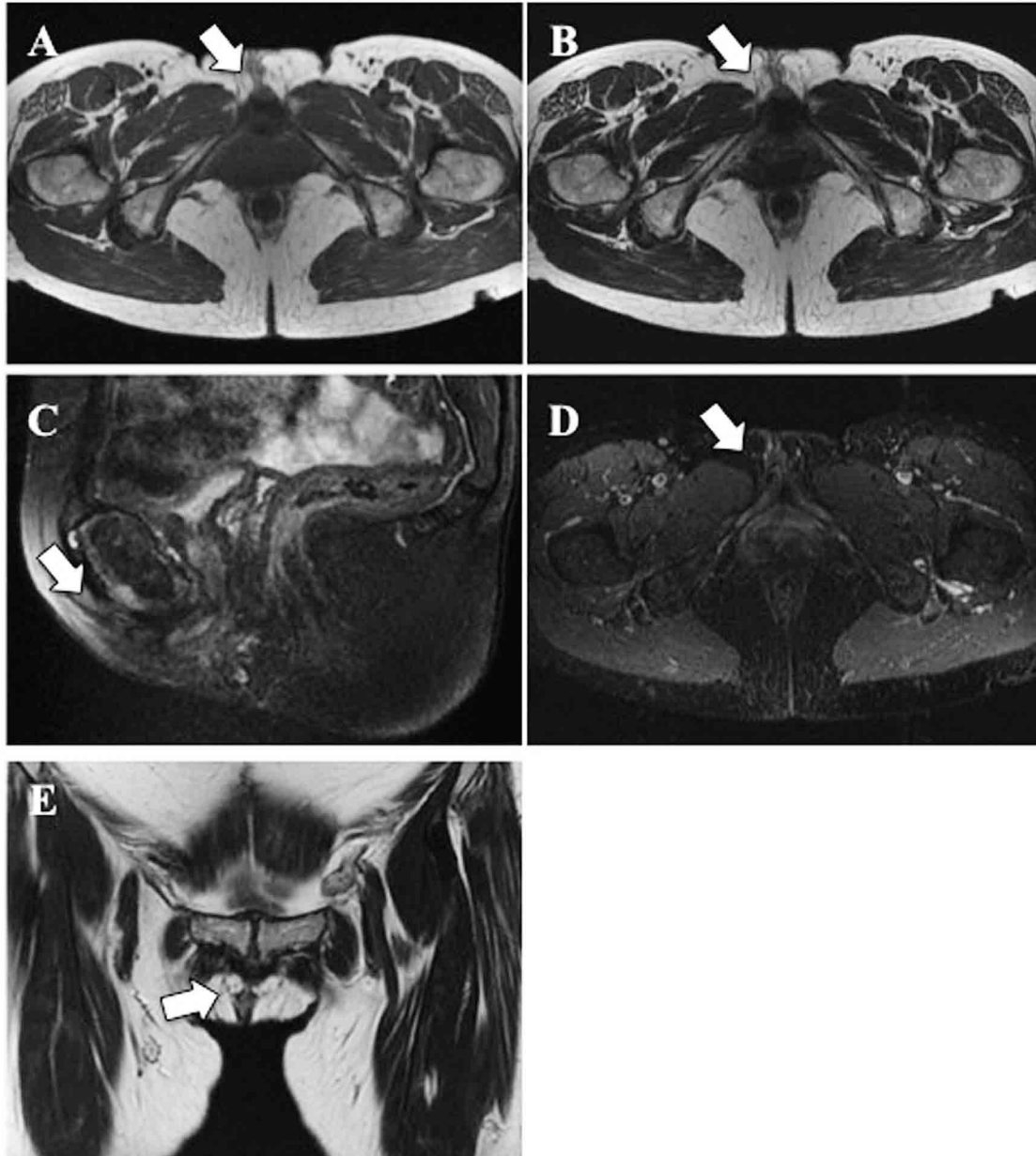


Figure 2 Follow-up MRI at 48 months

A : Axial T1-WI, B : axial T2-WI, C : sagittal fat-suppressed T2-weighted image, D : axial STIR, and E : coronal T2-WI images show the mass had disappeared completely

MRI, magnetic resonance imaging ; WI, weighted imaging ; STIR, short T1 inversion recovery

steroids resulted in only a temporary reduction of the mass and not in complete regression (7). Our patient had only dull pain and a pelvic brace may be effective because the pain and mass was disappeared after she wore the brace, so we opted for MRI follow-up of the lump.

In previous reports, watchful waiting was undertaken in 4 cases of subpubic cyst (2) ; cyst size did not change in 2 cases that were followed-up at 48 months or in one case that was followed-up at 9 months. Only one case showed slight reduction in size and improved symptoms on follow-up at 4 years. In comparison, the subpubic cyst in our case was relatively small, at $1.5 \times 1.0 \times 1.0$ cm. We speculate that size may influence spontaneous resolution of such benign cysts.

In conclusion, subpubic cartilaginous cyst may spontaneously regress. Although surgery would be necessary for severe symptomatic cases, in the case of asymptomatic or small subpubic cyst, observation and follow-up alone may be sufficient.

CONFLICTS OF INTEREST

The authors have no conflicts of interest to declare.

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Table 1 Clinical data for subpubic cartilaginous cyst

| Author (Year) | Case | Sex | Age | Gravida (G) and Para (P) | Symptoms | Size in MRI (mm) | Treatment | Follow-up result of observation | Complication |
|--|------|--------|----------------------|--------------------------|------------------------------|------------------------------------|---------------------------------|---------------------------------|--|
| Alguacil-Garcia <i>et al.</i> (1996) (1) | 1 | Female | Postmenopausal woman | NA | Pain and urinary dysfunction | NA | Operation | NA | NA |
| | 2 | Female | Postmenopausal woman | NA | Painless lump | NA | Operation | NA | NA |
| Kim <i>et al.</i> (2004) (5) | 3 | Female | 70 | G3P3 | Painless lump | 30 | Operation | NA | NA |
| Ergun <i>et al.</i> (2008) (2) | 4 | Female | 54 | G3 | Pain | NA | Observation after needle biopsy | No change after 48 months | NA |
| Bullock <i>et al.</i> (2009) (6) | 5 | Female | Postmenopausal woman | NA | Painless lump | 38 | Observation | No change after 48 months | NA |
| Hoogendoorn <i>et al.</i> (2009) (7) | 6 | Female | 55 | G4P3 | Pain | 37×36×35 | Operation | NA | Pain after operation |
| Judson <i>et al.</i> (2009) (8) | 7 | Female | 62 | G2 | Pain | 18×10×12 | Operation | NA | NA |
| Sava <i>et al.</i> (2012) (9) | 8 | Female | 59 | NA | Pain and urinary dysfunction | NA | Operation | NA | NA |
| Tan <i>et al.</i> (2012) (3) | 9 | Female | 69 | G4P4 | Painless lump | 50×30×30, 30×20×30 (multi-locular) | Observation after open biopsy | No change after 9 months | NA |
| Frag <i>et al.</i> (2014) (10) | 10 | Female | 61 | G3P3 | Urinary dysfunction | 32×30×39 | Operation | NA | Infection after operation |
| | 11 | Female | 56 | G1P1 | Urinary dysfunction | 30×38×27 | Operation | NA | infection and hematoma after operation |
| Wylie, K. R. (2014) (4) | 12 | Male | 69 | - | Pain and sexual function | 25 | Observation | Size reduction after 4 years | NA |

MRI=magnetic resonance imaging, NA=not available

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