A Rare Case of a Pelvic Digit Complicated by an Intramuscular Hematoma in a Patient with Severe Hemophilia B

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AUTHORS' CONTRIBUTIONS
Both Hae Weon Lee and Jessica Miller have written the manuscript and read and approved the final manuscript.

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DISCLOSURES
None

CONSENT
No

HUMAN AND ANIMAL RIGHTS
N/A

ABSTRACT

A pelvic digit, also described as a pelvic rib, sacral rib, or eleventh finger, is a rare congenital anomaly of ectopic bone adjacent to normal skeletal bone. Pelvic digits are usually unilateral and asymptomatic, with surgery only considered for symptomatic pelvic digits. Here, we present the first reported case in literature of a pelvic digit complicated by an intramuscular hematoma in the setting of severe hemophilia B.

CASE REPORT

BACKGROUND

To date, there have been no reported cases of a pelvic digit complicated by an intramuscular hematoma. This case illustrates the symptoms associated with the pelvic digit and intramuscular hematoma and highlights the importance of including pelvic digits in the differential diagnosis for back and lower extremity pain.

CASE REPORT

A 28-year-old male with history of severe hemophilia B (baseline factor activity < 1%) with recurrent hemarthrosis complicated by hemophilic arthropathy requiring multiple admissions presented to the Emergency Department with 1 week of atraumatic subacute back pain that radiated to his knee. The pain was described as constant, sharp and shocking, and exacerbated by muscle movement. No known inciting incident or associated symptoms such as fever, chills, or changes in skin color were reported.

On examination, the patient was visibly uncomfortable with tenderness to palpation of the right lateral buttock and thigh extending to the knee. Some fluctuance was noted about the proximal thigh. Laboratory workup was significant for mild anemia with hemoglobin of 13.6 gm/dL and partial thromboplastin time elevation to 103 seconds.

Anteroposterior and frog-leg lateral radiographs of the right hip demonstrated an osseous excrescence extending medially from the intertrochanteric femur (Figure 1). Initial interpretation was concerning for heterotopic ossification versus a congenital anomaly, with cross sectional imaging recommended for further evaluation. Computed Tomography (CT) (Figure 2) demonstrated a 5.4 cm corticated osseous structure located between the right proximal femur and ischial tuberosity, with the appearance suggestive of a pelvic digit. Extending along the pelvic digit was a large 8.7 cm intramuscular hematoma with multiple adjacent blood vessels (Figure 3). Although the inciting cause of the hematoma could not be confirmed, it was

hypothesized that mechanical impaction of the pelvic digit on the adjacent blood vessels was a contributing factor.

The patient was admitted to the Hematology service and managed with Factor IX and supportive measures with rest and ice. Orthopedic surgery was consulted for consideration of surgical removal of the pelvic digit; however, surgery was deferred given the associated risks and improvement with conservative management. The hematoma remained clinically stable, requiring no additional imaging, and he was ultimately discharged after 3 days with follow-up scheduled at the hematology treatment center. The patient was advised to return to the Emergency Department if there was any concern for recurrent bleeding prior to establishing care at the hematology treatment center.

On follow-up, the patient was initiated on 100 IU/kg of Factor IX once weekly to prevent bleeds and maintain current joint health. A bleed plan was also established, with additional Factor IX dosing parameters for mild to moderate bleeds and for severe bleeding. The patient reported resolution of the pain and swelling of his hip 2 weeks after discharge with regular prophylactic treatment.

DISCUSSION

Etiology & demographics

The pelvic digit was first documented by Sullivan and Cornwell in 1974 in a 15-year-old female with only a few additional cases reported thereafter [1]. While most commonly related to the ilium, the pelvic digit has also been found at the sacrum, coccyx, anterior abdominal wall, or pubic symphysis [2]. The exact incidence is unknown, but there were less than 20 reported cases as of the year 2000 [3].

Pelvic digits are usually unilateral, asymptomatic and found incidentally with only a few cases of bilateral or symptomatic pelvic digits, often located in the hip [4-9]. The etiology of the pelvic digit is not well established, but the proposed hypothesis is that this anomaly occurs from failure to fuse the costal primordium of the first coccygeal vertebra with the vertebral column within the first 6 weeks of embryogenesis [10]. Interestingly, this patient also presented with other congenital skeletal variations such as lumbosacral transitional vertebrae, however thus far, no correlation has been established between spinal congenital anomalies and pelvic digits.

Clinical & imaging findings

On plain radiographs, the pelvic digit has a characteristic phalanges- or rib-like appearance with variable pseudo-articulation [11]. On Computed Tomography (CT), the pelvic digit is composed of well-differentiated lamellar bone with the cortico-medullary pattern seen in skeletal bone.

Computed Tomography (CT) can be used to better localize and differentiate the pelvic digit from other differential diagnoses, helping to distinguish it from osteochondromas, myositis ossificans, or other causes of heterotopic bone formation.

Treatment & prognosis

The pelvic digit is considered a benign congenital anomaly that commonly is asymptomatic. It is important to note that pelvic digits are considered a “do-not-touch” lesion as further diagnostic tests can lead to unnecessary surgeries. Surgery is only considered for symptomatic pelvic digits, such as pelvic pain or back pain, that is refractory to conservative management.

For patients with hemophilia, it is integral for patients to receive comprehensive discharge instructions and precautions. The patient should be provided with education on how to administer prophylactic treatment, how to recognize signs and symptoms of future bleeds or compartment syndrome, medications to avoid such as non-steroidal anti-inflammatory drugs (NSAIDs) or other anticoagulants, and a plan on managing future bleeds such as returning to the Emergency Department or contacting their hemophilia treatment center. A follow-up appointment with a hemophilia treatment center or hematologist should be made prior to discharge.

Differential diagnoses

When diagnosing pelvic digits, it is important to consider other causes of ossified structures in the pelvis such as osteochondroma, myositis ossificans, ligamentous calcifications, and calcific tendinosis. Osteochondroma can be radiographically distinguished from a pelvic digit by the presence of a cartilaginous cap with endochondral ossification. Myositis ossificans demonstrates a fibroblastic core with peripheral bone and lacks pseudo-articulation which is often seen in pelvic digits. Calcifications in ligaments and tendons are often secondary to calcium deposition, amorphous in appearance, and lack the organization seen with mature bone [5].

In addition to radiographic findings, the location of these structures can be used to guide most likely diagnoses. Osteochondromas are commonly located at the knee, distal femur, and proximal tibia [12,13]. Myositis ossificans is often found at high-risk sites of injury such as the quadriceps femoris and brachialis muscle [14]. Ligamentous calcifications and calcific tendinosis occur most frequently at the rotator cuff, with the supraspinatus tendon being most involved [15,16].

An important differential consideration is heterotopic ossification (HO), which consists of zonal mineralization and peripheral mature cortical bone, and has been rarely described in the setting of hemophilia [17-20]. In addition to the classic findings of hemophilic arthropathy, this patient has evidence of heterotopic ossification (HO) at the elbow and knee presenting as small, calcified bodies and bulky osseous protuberances. Heterotopic ossification in the pelvis could conceivably mimic a pelvic digit and was considered in our patient; however, the

imaging appearance with presence of pseudo-articulations more closely resembled that of a pelvic digit. Although somewhat of a diagnostic dilemma, differentiating between the entities is likely entirely academic as it would not ultimately change management.

TEACHING POINT

The pelvic digit, also known as pelvic rib, sacral rib, or eleventh finger, is a rare congenital anomaly that most commonly arises from the ilium, is asymptomatic, and often does not require treatment. It is considered a "do-not-touch" lesion; therefore, surgery is only considered for symptomatic pelvic digits refractory to conservative management.

QUESTIONS

Question 1: Which pathology causing ossification of structures presents radiographically with presence of a cartilaginous cap with endochondral ossification?

1. Pelvic Digit
2. Osteochondroma (applies)
3. Myositis ossificans
4. Calcific tendinosis
5. Ligamentous calcifications

Explanation:

1. Pelvic digits have phalanges- or rib-like appearance with a cortico-medullary pattern seen in skeletal bone [On plain radiographs, the pelvic digit has a characteristic phalanges- or rib-like appearance with variable pseudo-articulation].
2. The cartilaginous cap with endochondral ossification is the classic appearance of osteochondromas on plain radiographs [Osteochondroma can be radiographically distinguished from a pelvic digit by the presence of a cartilaginous cap with endochondral ossification.]
3. This appears as a fibroblastic core with peripheral bone that lacks pseudo-articulation [Myositis ossificans demonstrates a fibroblastic core with peripheral bone and lacks pseudo-articulation which is often seen in pelvic digits]
4. This would appear amorphous in appearance without the organization seen with mature bone. [Calcifications in ligaments and tendons are often secondary to calcium deposition, amorphous in appearance, and lack the organization seen with mature bone]
5. This would appear amorphous in appearance without the organization seen with mature bone. [Calcifications in ligaments and tendons are often secondary to calcium deposition, amorphous in appearance, and lack the organization seen with mature bone]

Question 2: Where does the pelvic rib most commonly arise from?

1. Sacrum
2. Coccyx
3. Pubic symphysis
4. Abdomen
5. Ilium (applies)

Explanation: This congenital anomaly has most frequently been reported at the ilium and is usually unilateral. [While most commonly related to the ilium, the pelvic digit has also been found at the sacrum, coccyx, anterior abdominal wall, or pubic symphysis.]

Question 3: What is an important differential diagnosis that can result from repeated bleeding in patients with hemophilia?

1. Osteochondroma
2. Myositis ossificans
3. Heterotopic ossification (applies)
4. Calcific tendinosis
5. Ligamentous calcifications

Explanation: In patients with hemophilia, such as the patient in this case, recurrent bleeding and damage to surrounding structures can lead to heterotopic ossification. This should always be considered in the differential diagnosis of pelvic digits in this patient population. [An important differential consideration is heterotopic ossification (HO), which does consist of well-differentiated bone, and has been rarely described in the setting of hemophilia.]

Question 4: How do you manage pelvic digits that are asymptomatic?

1. No treatment needed (applies)
2. Annual re-imaging
3. Pain relievers
4. Biopsy for further evaluation
5. Surgical removal

Explanation:

1. As pelvic digits are considered a "do-not-touch" lesion, asymptomatic patients do not require any intervention. [It is important to note that pelvic digits are considered a "do-not-touch" lesion as further diagnostic tests can lead to unnecessary surgeries.]
2. Pelvic digits are benign and do not require repeated follow-up unless symptomatic. [It is important to note that pelvic digits are considered a "do-not-touch" lesion as further diagnostic tests can lead to unnecessary surgeries.]
3. Conservative management with pain relievers can be utilized for symptomatic, not asymptomatic, patients. [Surgery is only considered for symptomatic pelvic digits, such as pelvic pain or back pain, that is refractory to conservative management with pain relievers.]
4. Asymptomatic pelvic digits should not have further diagnostic tests after radiographic identification. [It is important to note that pelvic digits are considered a "do-not-touch" lesion as further diagnostic tests can lead to unnecessary surgeries.]
5. This is only reserved for patients whose symptoms are not controlled with conservative management. [Surgery is only considered for symptomatic pelvic digits, such as pelvic pain or back pain, that is refractory to conservative management with pain relievers.]

Question 5: What imaging modality can be used to further characterize and differentiate pelvic digits from other types of ossification?

1. X-ray
2. Magnetic Resonance Imaging (MRI)
3. Nuclear Imaging
4. Computed Tomography (CT) (applies)
5. Ultrasound

Explanation: Other causes of ossification to consider when diagnosing pelvic digits are osteochondromas, myositis ossificans, ligamentous calcification, or calcific tendinosis. These differential diagnoses can be distinguished from each other through computed tomography. [On CT, the pelvic digit is composed of well-differentiated lamellar bone with the cortico-medullary pattern seen in skeletal bone. CT can be used to better localize and differentiate the pelvic digit from other differential diagnoses.]

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FIGURES

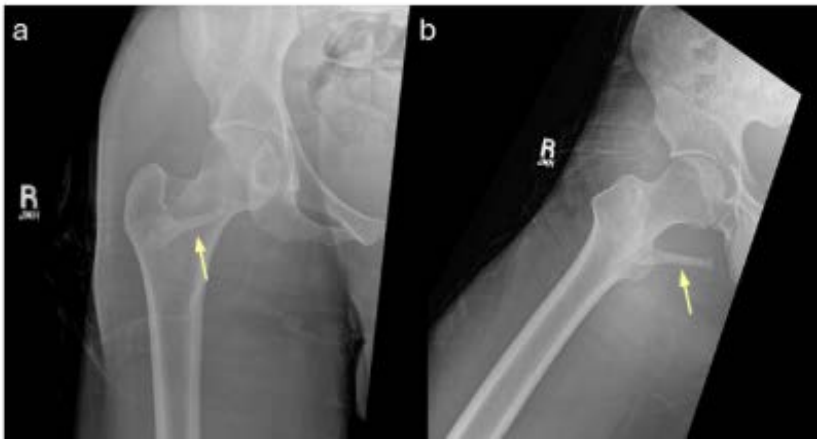

Figure 1: 28 year old male with right sided pelvic digit.

FINDINGS: AP and frog-leg lateral plain radiograph of the right hip demonstrates an osseous excrescence (yellow arrows) extending between the intertrochanteric right proximal femur and ischial tuberosity. No periosteal reaction or acute fracture. Normal appearance of the hip joint.

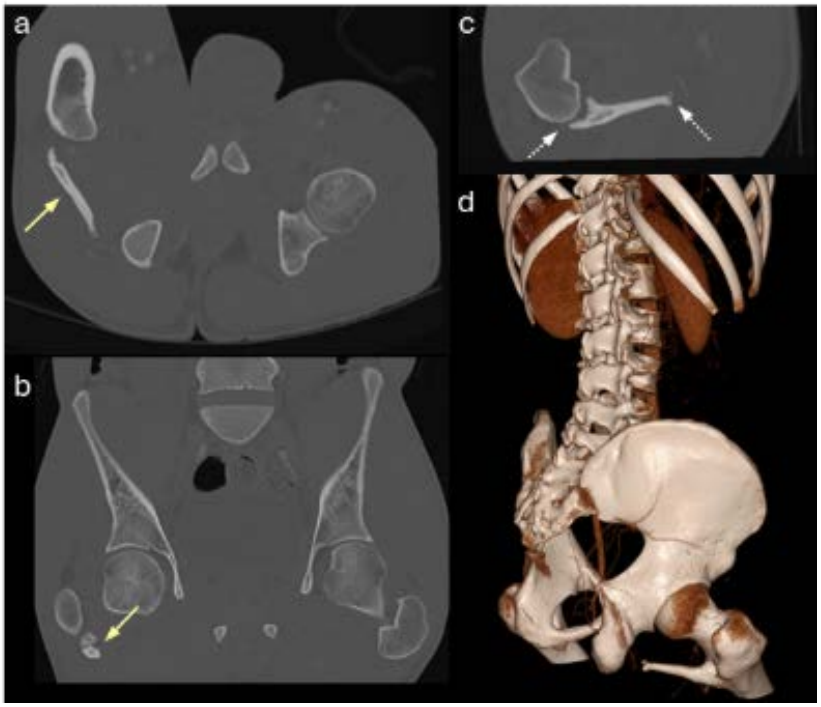

Figure 2: 28 year old male with right sided pelvic digit.

FINDINGS: Contrast-enhanced Computed Tomography (CT) with reconstructed images demonstrating the pelvic digit (yellow arrows) measuring 5.4 x 0.7 x 1.8 cm (L x W x H) extending between the right proximal femur and ischial tuberosity. Pseudo-articulations are present proximally and distally (dashed arrows).

TECHNIQUE:

a) Axial, b) Coronal, c) Sagittal Computed Tomography (CT) with total mAs 801, 120kV, 1mm slice thickness, 100mL iohexol (Omnipaque) 350mg/mL obtained from lung bases through the proximal femora following the administration of contrast material

d) Three-dimensional reconstruction with pelvic digit and adjacent pelvic vasculature

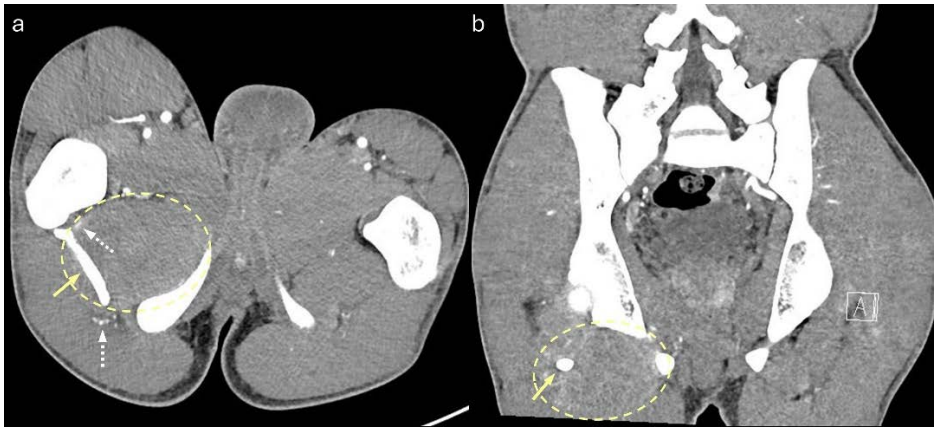

Figure 3: 28 year old male with right sided pelvic digit complicated by intramuscular hematoma.

FINDINGS: Contrast-enhanced Computed Tomography (CT) demonstrating an intramuscular hematoma measuring 8.7 x 6.3 x 5.5 cm (L x W x H) within the proximal right thigh (encircled with dashed line). The hematoma is adjacent to the pelvic digit (yellow arrow), which is near multiple blood vessels (dashed arrows).

TECHNIQUE: a) Axial, b) Coronal Computed Tomography (CT) with total mAs 801, 120kV, 1mm slice thickness, 100mL iohexol (Omnipaque) 350mg/mL obtained from lung bases through the proximal femora following the administration of contrast material

Table 1: Summary table of pelvic digits

Etiology	Failure to fuse the costal primordium of the first coccygeal vertebra with the vertebral column within the first 6 weeks of embryogenesis
Incidence	Rare As of the year 2000, only 20 reported prior cases
Imaging Findings	Plain Radiograph: phalanges- or rib-like appearance with variable pseudo-articulation Computed tomography (CT): well-differentiated lamellar bone with the cortico-medullary pattern seen in skeletal bone
Treatment	Conservative management (i.e. pain relievers) – “do-not-touch” lesion Surgery only reserved for refractory symptoms
Prognosis	Usually asymptomatic with excellent prognosis. Does not require follow-up.

Table 2: Differential diagnosis table of pelvic digits

Diagnosis	**Most Common Location(s)**	**Imaging Findings**
Pelvic digit	Ilium	Plain radiograph: phalange- or rib-like appearance with variable pseudo-articulation Computed Tomography (CT): well-differentiated lamellar bone with the cortico-medullary pattern seen in skeletal bone
Osteochondroma	Knee Distal femur Proximal tibia	Plain radiograph: sessile or pedunculated appearance seen in metaphyseal region, commonly projecting away from epiphysis with variable appearance of cartilaginous cap Computed Tomography (CT): cartilaginous cap with endochondral ossification
Myositis ossificans	Quadriceps femoris Brachialis muscle	Plain radiograph: calcification with classic well-circumscribed peripherally calcified appearance by 2 months Computed Tomography (CT): fibroblastic core with peripheral bone lacking pseudo-articulation
Ligamentous calcification/ Calcific tendinosis	Supraspinatus tendon (rotator cuff)	Plain radiograph: homogeneous hyperdensity with variable morphology, commonly with globular/amorphous appearance Computed Tomography (CT): amorphous in appearance lacking the organization seen with mature bone
Heterotopic ossification	Thigh Pelvis	Plain radiograph: peripheral calcification that evolve from poorly organized appearance to mature cortical bone Computed Tomography (CT): zonal mineralization and peripheral mature cortical bone

KEYWORDS

Pelvic digit, pelvic rib, sacral rib, eleventh finger, hematoma, hemophilia B