Author's response to reviews

Title: Isolated loss of inferior pubic ramus: A case report.

Authors:

Aly Saber (aly­saber@­ho­tmail.com)

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Isolated loss of inferior pubic ramus: 
A case report

Aly Saber, MD
Consultant Surgeon
Port-Fouad General Hospital

Mobile: +2/ 0123752032
Home: +2/066/3406474
Clinic: +2/066/3236922

Correspondence:____

Dr/Aly Saber
Port-Fouad General Hospital
Port-Fouad, Port-Said, Egypt

alysaber@hotmail.com
Abstract:

Introduction:
It was stated that regulation of iliac bone development is
different from that of ischium and pubis. There are well-known
clinical syndromes concerning with hypoplasia of ischiopubic bone;
small patella syndrome, nail-patella syndrome, Ischio-pubic-
patellar hypoplasia and Ischiopubic hypoplasia

Case presentation:
A young adult female, 35 years old with normal daily activity,
presented with pain in the left lower limb since 6 months. She
sought advice with her orthopedic surgeon and referred to the
author to exclude primary soft tissue neoplasm.

There was no history of trauma, chronic medical illness, or
surgical operations. Full systemic examination showed no soft
tissue swelling or any other bony defects. Laboratory
investigations and whole body imaging were performed and
showed also the same.

Isolated loss of the left inferior pubic ramus and thinned out
the superior were detected raising following questions if the lesion
is secondary osteolytic lesion, endocrine disease or primary
osteolytic lesion.

Conclusion:
Isolated loss of inferior pubic ramus with no concomitant
bony or soft tissue anomalies is still unknown uptill now. To the
author's knowledge, this finding has not been described previously
Introduction:

The development of the pelvic girdles has been poorly investigated and reported evidences suggested that the regulation of ilium development is different from the development of ischium and pubis (1).

An extensive work was done to study the prenatal development and mineralization of ossification centres in the pelvic bone (ilium, ischium, and pubic bone) using radiography and optical density measurements on human fetuses. The mineral density of the pelvic bone increases with age and the mineralization rate changes throughout fetal life (2).

There are well-known clinical syndromes concerning with hypoplasia of ischiopubic bone; small patella syndrome, nail-patella syndrome, Ischio-pubic-patellar hypoplasia and Ischiopubic hypoplasia [3].

Case Presentation:

A young adult female, 35 years old with normal daily activity, presented with pain in the left lower limb since 6 months. She sought advice with her orthopedic surgeon and referred to the author to exclude primary soft tissue neoplasm.

She denied any history of trauma, violence or abnormal muscular overload. There was no history of chronic medical
illness, or surgical operations. There was no family history of congenital defects or similar conditions.

On examination, she was apparently well-built, and full systemic examination showed no soft tissue swelling or any other bony defects with special effort to both breasts and thyroid. There was no evidence of any congenital anomalies especially the genitalia, hip bones, long bones of lower limbs or chest.

Laboratory investigations were performed and showed normal organ functions. Serum calcium and phosphorus were estimated as well as the parathyroid hormone assay. All proved normal values.

Whole body imaging started with plain x-ray films showed isolated loss of the left inferior pubic ramus and thinned out the superior with no other bony anomalies (fig.1). CT cuts also showed the same data with normal muscular attachment (fig.2a) and the thin left superior ramus was shown in other cut (fig.2b). The same data were obtained from MRI films.

Bone scan was performed and excluded any osteolytic lesion, bone rarefaction, cysts or neoplasm (fig.3).

Isolated loss of the left inferior pubic ramus and thinned out the superior were detected and the following questions were put if the lesion is secondary osteolytic lesion, endocrine disease or primary osteolytic lesion.
**Discussion:**

Study of human limb malformation syndromes revealed some clinical syndromes concerning with hypoplasia of ischiopubic bone; small patella syndrome, nail-patella syndrome, Ischio-pubic-patellar hypoplasia and Ischiopubic hypoplasia. All described pubic bone defect as well as other bony or soft tissue anomalies [3].

Small patella syndrome (SPS) is characterized by patellar aplasia or hypoplasia and by anomalies of the pelvis and feet, including disrupted ossification of the ischia and inferior pubic rami [4].

Bilateral absence of the patella in an 11-year-old girl with absence of the ischial and inferior pubic rami bilaterally together with skeletal and soft-tissue deformities are reported and may represent a unique syndrome [5].

Hypoplasia of the ischiopubic region together with spinal dysraphism and scoliosis as well as bilateral aplasia of the patella is an extremely rare anomaly [6].

Genitopatellar syndrome is a newly described disorder characterized by absent/hypoplastic patellae, lower extremity contractures, urogenital anomalies, dysmorphic features, skeletal anomalies, and agenesis of the corpus callosum [7].
Unilateral hip dislocation in conjunction with ipsilateral absence of the pubic bone, an undescended palpable testicle and hypospadias collectively form a syndrome has not been reported in the orthopaedic literature previously [8].

All these clinical syndromes showed multiple bony and soft tissue anomalies [3-8], but in our case there is isolated loss of inferior pubic ramus without any concomitant bony or soft tissue anomalies. Also, it is not associated with genital anomalies. To author’s knowledge, this case is not described uptill now through world-wide search.

The patient of this case report remained free from complaint for 35 years and this notice may be matched with reported data of patients aged from 20 to 70 years. The main complaint at consultation was with their knees due to patellar instability and pain [9].

Many patients were presented early in their lives, but lack of significant clinical complaints were also reported in a 77-year-old woman with nail patella syndrome [10].

**Conclusion:**

Hypoplasia of the ischiopubic region is described in some syndromes together with other bony and soft tissue anomalies. But isolated loss of inferior pubic ramus without any concomitant bony or soft tissue anomalies uptill now is still unknown to the author's knowledge according to the world wide search.
The author put the following questions to solve the problem. If this lesion is congenital, why is it isolated with no other visceral or bony manifestations? It may be secondary to eroding traumatic haematoma or vascular insult.

**Consent:**

The author stated that written informed patient consent was obtained for publication of the report on The Journal of Medical Case Reports as well as accompanying images.

**Competing interests**

The author declares that he has no competing interests.

**Author's contributions:**

The author examined the patient, requested the performed investigations, initiated drafting of manuscript, prepared the figures and got acquisition of data through web.

**Acknowledgements**

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References:


Figure legends:

Figure 1: plain x-ray films showed isolated loss of the left inferior pubic ramus and thinned out the superior with no other bony anomalies.

Figure 2: CT cuts also showed the same data with normal muscular attachment (fig.2a) and the thin left superior ramus was shown in other cut (fig.2b).

Figure 3: Bone scan was performed and excluded any osteolytic lesion, bone rarefaction, cysts or neoplasm.