

Unveiling the Rarity: A Case Report of Rudimentary Third Lower Limb

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Abstract

Extra supernumerary lower limb a very rare manifestation in human being and animals. Till date literature review reveals around 31 cases. We are presenting such a case on Pygomelia of a 3 month old baby with a rudimentary (third) lower limb arising from her right buttock. Clinical examination revealed an extra lower limb that was arising from para-medial aspect of the right gluteal region just lateral to the gluteal cleft. The limb had some degrees of movement at the hip and rudimentary knee. Radiographically, the extra limb had a compete femora with a rudimentary tibia, ankle and foot. The knee and ankle joints were not well formed. After giving a racquet-shaped incision, two neuro-vascular bundles were identified at the base of the limb that were ligated and transected. The limb was disarticulated from its pseudo-joint. Post-operative recovery was uneventful with intact neurovascular status of both normal limbs. The wound seemed well healed at 4 weeks follow-up. Surgical resection of the supernumerary extremity is advocated in patients having accessory limbs that look odd and bear a significant psycho-social impact on the patient and their parents. The complete removal of the limb from its root along with the epiphysis associated with its pseudo-joint is necessary to prevent regrowth of the femur from the left over physis.

Keywords: Pygomelia; Supernumerary; Extra; Disarticulation; Regrowth.

Introduction

Extra supernumerary lower limb a very rare manifestation in human being and animals. Till date literature review reveals around 31 cases [1, 2, 3, 4, 5]. It has a heterogenous pathogenesis [1] which may occur as a complete or incomplete limb. Such limbs arising from gluteal region are named as Pygomelia [2]. The limb often looks like an incomplete tail of a monkey with some range of motion and power depending on how well the joints are formed. We are presenting such a case on Pygomelia arising from the right buttock with intact sensations but no motor innervation.

Case Summary

A 3 months old baby presented with a rudimentary (third) lower limb arising from her right buttock. She was born of non-consanguineous marriage with no family history for musculoskeletal abnormalities. The mother's pre-natal and post-natal history was un-eventful. There was no history of drug use or viral infections etc. The baby has achieved timely developmental milestones till date, except an odd-looking extra lower limb that hampers the baby's perineal hygiene care and is a cause of the parents' psychological upset.

Clinical Examination

Revealed an extra lower limb that was arising from para-medial aspect of the right gluteal region just lateral to the gluteal cleft. The limb had some degrees of movement at the hip and rudimentary knee. The foot was small, and incompletely formed with three digits. The limb was fusiform in shape with greater girth proximally as compared to distal. A skin dimple was noted in the distal half of the limb overlying the knee joint. Only a few degrees of passive movement could be elicited at the knee joint. A second crease was noted at the distal third, overlying the ankle joint with no passive movement. The foot had two clefts and three toes [Figure 1-C].



Figure 1: Clinical images showing rudimentary third limb arising from the postero-medial aspect of right gluteal region. Subset (lower right) showing the incompletely formed foot with two clefts and 3 toes.

Radiographic Evaluation

Revealed a normal right lower limb along with an extra third lower limb arising from the medial aspect of right hip joint. The extra limb had a complete femora with a rudimentary tibia, ankle and foot. However, the knee and ankle joints were not well formed. [Figure 2].

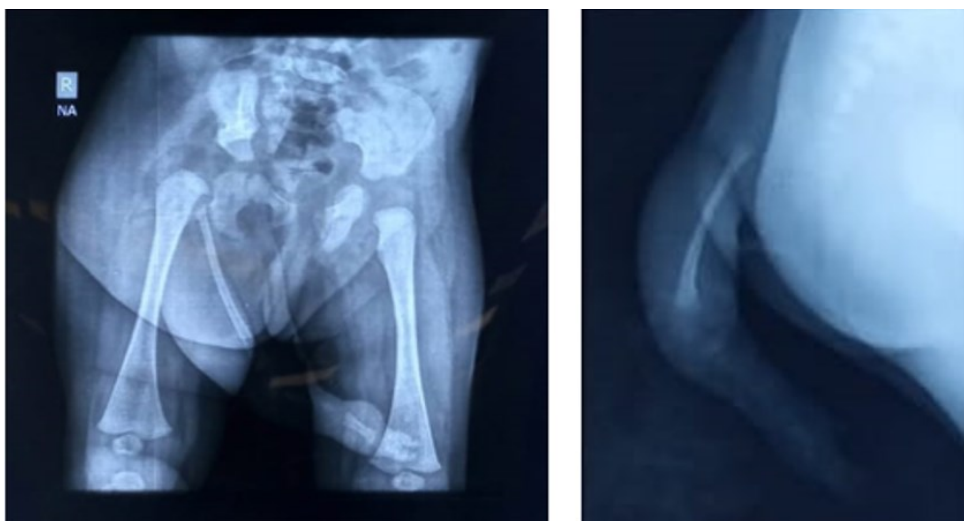


Figure 2: Radiographs showing bony constituents of the accessory limb including femur, tibia (segmented) and foot.

Surgical Steps

A Racquet shaped incision was made at base of the rudimentary limb. A fascio-cutaneous flap was raised, Sharp deep dissection was continued to root of the limb arising from gluteal region. Two neuro-vascular bundles were identified at the base of the limb that were ligated and transected. Both femoral nerve and artery, and sciatic nerve were well formed. The limb was disarticulated from the its pseudo-joint positioned inferomedial to original hip joint. [Figures 3A & 3B] show complete resection of the limb with well-formed femoral head. Haemostasias secured and wound closed in layers [Figure 3C]. The post-operative recovery was uneventful with intact neurovascular status of both normal limbs. At 2- & 4-weeks' follow-up the wound was healthy with progressive healing and acceptable cosmetic look and happy note of parents.

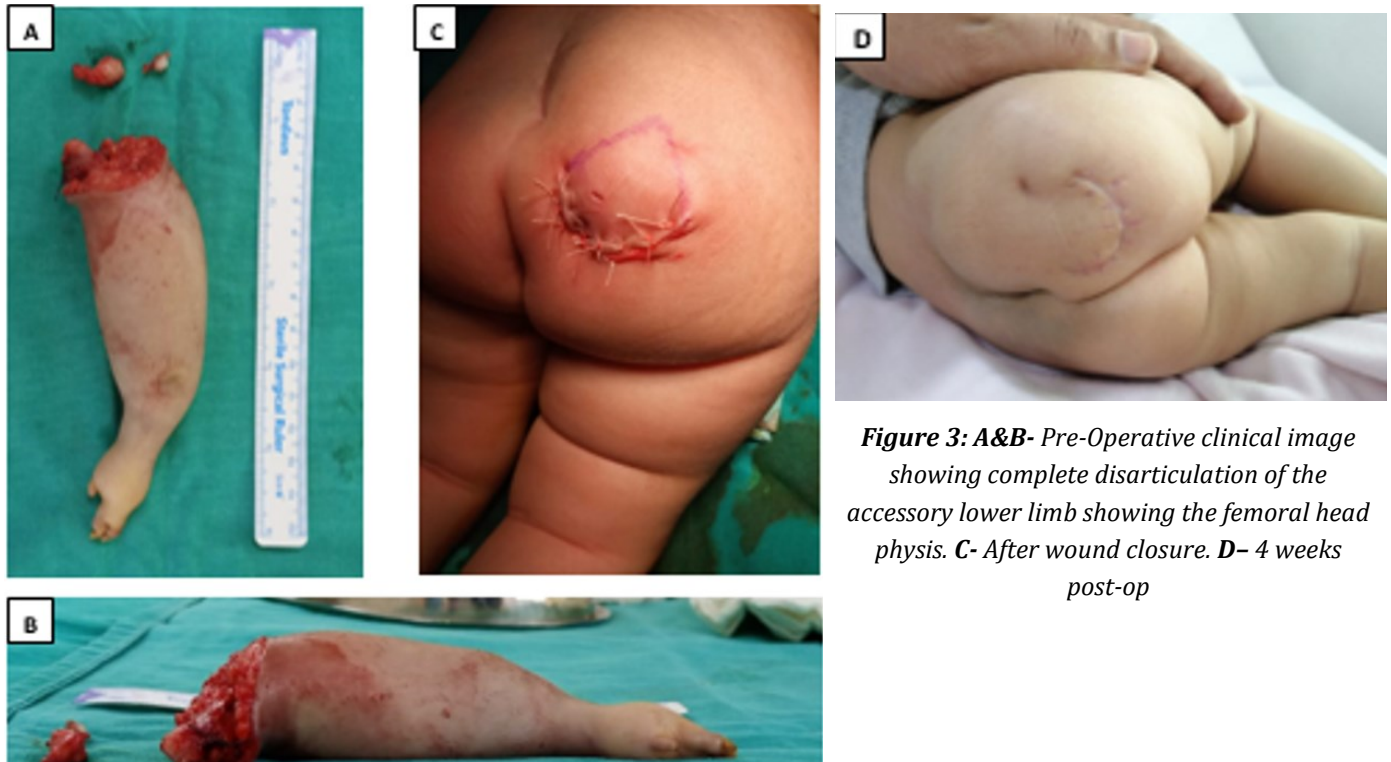


Figure 3: *A&B- Pre-Operative clinical image showing complete disarticulation of the accessory lower limb showing the femoral head physis. C- After wound closure. D- 4 weeks post-op*

Discussion

Limb development is a complicated embryological process which involves multiple genes and demands precise gene regulation for a normal growth process [1]. Errors in formation may include bifurcation or splitting of the limb bud (Hanley) [6], error of limb bud specification (Packard) [7], or a flaw in the genetic mechanism [8] resulting in duplication [5]. Based on its the part of the body, it is attached to, it can be classified as Cephalomelia (attached to the head), Notomelia (attached to the backbone), Thoracomelia (Attached to the thorax), and Pryomelia (Attached to the pelvis) [2]. Surgical resection of the supernumerary extremity is advocated in patients having accessory limbs [1] that look odd and bear a significant psycho-social impact on the patient and their parents. The complete removal of the limb from its root along with the epiphysis associated with its pseudo-joint is necessary to prevent regrowth of the femur from the left over physis [Figure 3A].

Our case amazingly had a completely formed hip joint along with a separate neuro-vascular bundle (The sciatic nerve branching from the original sciatic nerve; however, the femoral nerve bundle's origin could not be confirmed). We disarticulated the limb from its pseudo joint, completely removing the femoral head (including physis) in order to prevent its re-growth.

Conclusion

We presented the case of tail like duplication of the lower limb. Early diagnosis (pre-natal ultrasonography) can facilitate effective management and address psychological concerns of the parents at birth. Complete disarticulation is mandatory prevent re-growth from left-over physis.

Conflict of Interest

The authors declare that they have no conflict of interest.

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