



## PYGOMELIA AND TRUE HERMAPHRODITISM IN A NINE WEEK OLD LARGE WHITE PIGLET CASE REPORT

Ajadi, T. A.<sup>1</sup>, Olaniyi, M. O.<sup>2</sup>

<sup>1</sup>Department of Veterinary Public Health and Reproduction

<sup>2</sup>Department of Veterinary Pathology, Federal University of Agriculture, Abeokuta  
Ogun State  
Nigeria

ayisat\_ajadi@yahoo.com

### ABSTRACT

A nine weeks old female Large White piglet which was presented to the Veterinary Teaching Hospital, Federal University of Agriculture, Abeokuta, with a complaint of extra limbs was diagnosed with pygomelia and concurrent true hermaphroditism based on gross morphologic features, radiography, exploratory laparotomy and histopathology of the malformed organs. The piglet had two well-developed extra hind limbs consisting of the femur, tibia, fibula and the phalanges. Radiographically, the accessory limbs were attached to the ischium through a rudimentary pelvic bone. The supernumerary limbs were smaller than the normal appendages, but contained equal digits. The anal orifice was observed cranial to the right supernumerary limb. Caudal to the left supernumerary limb a rudimentary penis was observed. Two oval shaped fibrous masses were palpated in the inguinal canal of the piglet. In addition, there was a transparent tubular tract measuring 24 cm in length which contained serous fluid. The right kidney was rudimentary measuring 2.10 cm, while the left kidney appeared

hypertrophied measuring 6.10 cm. The histology of the left kidney showed dysplastic areas of undifferentiated mesenchymal stroma in the cortex and medulla with the presence of groups of immature glomeruli in the cortex. The tubules in the medulla were scanty in number and had atypical epithelium. The adrenal glands had normal architecture with ectopic adrenal tissue in the adrenal capsule, while the ovaries and uterus were normal. It was concluded that the complex anomalies in the piglet might be as a result of a complex mode of inheritance.

**Key words:** hermaphroditism; intersex; pigs; pygomelia

### INTRODUCTION

Developmental malformations occur relatively frequently in swine in comparison to other domestic species. Frequently reported disorders include myofibrillar dysplasia, umbilical and inguinal hernias, cryptorchidism, intersexes, and anal atresia [3]. The overall incidence of develop-

mental defects in piglets was 2.07 % [14]. The frequency of developmental anomalies in pigs depends upon the breed and population, and has been reported to be decreasing in some countries [2].

Congenital anomalies may be caused by genetic factors (transgenes, chromosomes), environmental agents (infections, teratogens, toxins, fertilization techniques, management) or a combination of factors [7]. There have also been reports indicating that chromosomal aberrations are associated with congenital limb malformations [9]. A polygenic mode of inheritance has been suggested in most of the cases, however, not all congenital defects are heritable.

The congenital anomalies of the musculoskeletal system are considered to be one of the most frequent anomalies found in humans and animals [8]. Supernumerary ectopic limbs or polymelia is a congenital anomaly in which accessory limbs are attached to the various body regions [4]. These accessory limbs are smaller than normal limbs, and are associated with stiff joints and sparse muscles without innervations. The condition has been reported in calves, ewes, goats, poultry and in humans, but they are extremely rare in swine [12, 13]. Only one case of polymelia has been described in swine and it was associated with features such as *duplicitas coli partialis et recti*, *atresia ani* with *recto-genital fistula*, *duplicitas corpori uteri*, *cervicis*, *vaginae et vulvae*, and *duplicitas vesicae, urethrae et renalis* [11].

Pygomelia is a variant of polymelia in which the accessory limbs are attached to the pelvis through rudimentary *os coxae*. It usually results from a caudal bifurcation of the long axis of the body. It has been reported in cross breed calves [6, 10]. However, no record of pygomelia in pigs could be found in the literature. This paper described a rare case of pygomelia in a nine weeks old female large white piglet. The congenital defect was also associated with a true hermaphroditism.

## CASE PRESENTATION

A nine weeks old female large white piglet was presented with a complaint of extra limbs to the Veterinary Teaching Hospital, Federal University of Agriculture, Abeokuta; from a farm of 40 adult pigs located at Ifo local government area of Ogun State in Nigeria. It was one of a litter of six piglets farrowed by a primiparous 16 months old sow mated to a 14 months old boar. The pigs on the farm were fed blood meal, palm kernel cake, cassava peels and water

leaves. An outbreak of African swine fever was reported on the farm in 2009, although the current pigs on the farm were from a new stock. There were no records of umbilical hernia or any other congenital anomaly from the farm except for the current case. The anomaly was detected at birth and did not affect the parturition in the sow.

The piglet weighed 3 kg at presentation, while the posture and conformation of the piglet reflected a healthy appearance. The skin was smooth with no abnormality. All the physiological parameters were essentially normal. An examination revealed two well-developed extra hind limbs (Fig. 1), consisting of the femur, tibia, fibula and the phalanges, which were attached around the mid pelvis caudal to the tail of the piglet. The supernumerary limbs were smaller than the normal appendages, but contained equal number of digits and the appendages were non-functional. The anal orifice was observed cranial to the right supernumerary limb, and caudal to it was a vagina, while a penis was observed caudal to the left supernumerary limb. Two oval shaped fibrous masses thought to be the testes were palpated in the inguinal canal of the piglet. Other external features of the piglet were essentially normal.

## MANAGEMENT AND OUTCOME

Antero-posterior and lateral radiographs of the abdomen and hip of the piglet were obtained using a mobile

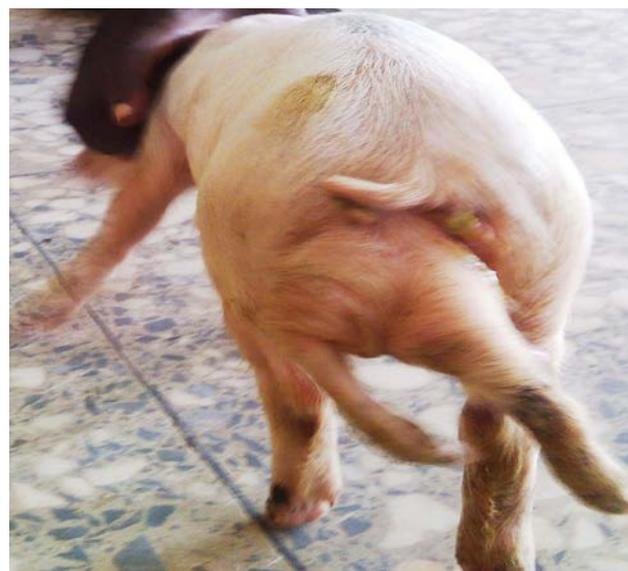


Fig. 1. Picture of a nine weeks old Large White piglet with supernumerary limb (SL)

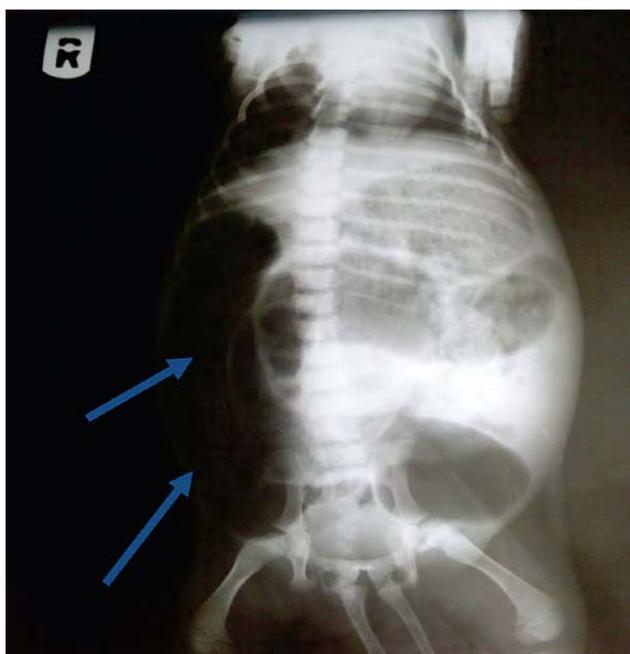


Fig. 2. Ventro-dorsal radiograph of the abdomen of an eight weeks old Large White piglet showing the gas filled caecum and colon



Fig. 3. Antero-posterior radiograph of eight weeks old piglet showing a rudimentary limb (RF) attached to the ischium (I) of the piglet. The main femur (F) is parallel to the rudimentary femur



Fig. 4. Caudo-dorsally located transparent tubular structure from the abdomen of an eight weeks old Large White piglet

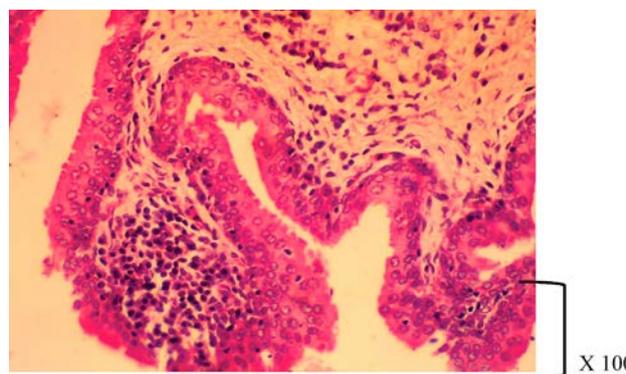


Fig. 5. Photomicrograph of a transparent caudo-dorsal abdominal tubular structure showing segmental hyperplasia of transitional epithelial mucosa and hyperplasia of associated lymphoid tissue. Magn.  $\times 100$

x-ray machine (PLH Medicals, Herts, UK). Thereafter, the piglet was anaesthetized using intramuscular injections of  $0.04 \text{ mg.kg}^{-1}$  of atropine sulphate (Atocan<sup>®</sup>, Sishui Xierkang Pharma, China),  $0.5 \text{ mg.kg}^{-1}$  of 2% xylazine (Xylazine 20 Inj<sup>®</sup>, Kepro, Holland),  $0.5 \text{ mg.kg}^{-1}$  diazepam (Calmpose<sup>®</sup>, Ranbaxy, Dewas, India) and  $10 \text{ mg.kg}^{-1}$  of 5% ketamine hydrochloride (Rotexmedica, Trittau, Germany). Two percent lignocaine was also infiltrated around the ventral mid-line to obtain good somatic analgesia. Thereafter, an exploratory laparotomy was done to check if there were other

anomalies internally. The structurally deformed abdominal organs were excised and fixed in 10% formaldehyde for histopathology.

The abdominal radiograph of the piglet revealed radio-lucent gas filled caecum and colon and increased soft tissue opacity around the middle and caudal abdominal cavity (Fig.2). There was a completely formed but rudimentary pelvic bone which attached the accessory limb to the ischium of the piglet (Fig.3). The colon and caecum were gas distended and occupied most of the part of the abdominal cavity. A transparent tubular tract measuring about 24 cm in length and containing serous fluid was observed cranio-dorsal to the bladder (Fig.4). The histology of the structure



Fig. 6. Kidneys of an eight weeks old Large White piglet showing a normal kidney (Big arrow) and a rudimentary kidney (Small arrow)

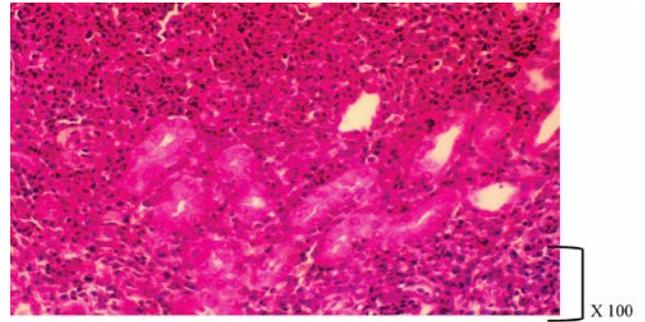


Fig. 7. Photomicrograph of the rudimentary kidney of the piglet showing dysplastic areas of undifferentiated mesenchyme in the cortex and medulla with the presence of immature blind ending tubules. Magn.  $\times 100$



Fig. 8. Paired rudimentary oval shaped structures measuring about 2.4 cm in length from the inguinal canal of the piglet

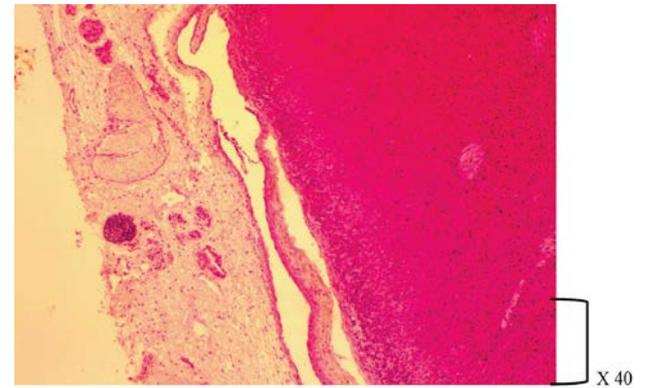


Fig. 9. Photomicrograph of the inguinal structure showing a normal architecture of the adrenal gland with ectopic medullary tissue in the cortex. Magn.  $\times 40$

revealed segmental hyperplasia of the transitional epithelium mucosa and the associated lymphoid tissue (Fig. 5). The ovaries and uterus were normal. The right kidney was rudimentary measuring 2.10 cm, while the left kidney appeared hypertrophied measuring 6.10 cm (Fig. 6). The histology of the left kidney showed dysplastic areas of undifferentiated mesenchymal stroma cells in the cortex and medulla with the presence of groups of immature glomeruli in the cortex (Fig. 7). The tubules in the medulla were scanty in number and had atypical epithelium. The spleen, liver and stomach of the piglet appeared essentially normal. An incision into the inguinal canal revealed two rudimentary oval shaped structures measuring about 2.4 cm in length (Fig. 8). The histology of the adrenal gland showed normal architecture with foci of ectopic parenchymal tissue in the thickened adrenal capsule (Fig. 9).

## DISCUSSION

Congenital defects like umbilical and inguinal hernias, cryptorchidism, intersexes, and anal atresia occur relatively frequently in pigs [3]. However, congenital defects of the limbs are very rare in pigs. Polymelia with dactylogryposis have been reported in a five months old female Landrace and Pietrain piglet cross [11]. There appears to be no report of congenital limb anomaly in the Large White pigs. This report describes a complex malformation in a Large White piglet characterized by accessory limbs, rudimentary left kidney, an accessory and rudimentary urinary system, and ectopic adrenal tissues located within the thickened adrenal capsule. In addition, the piglet was an intersex with an external penis, inguinally located testes with bulbourethral gland and well-formed ovary, uterus and vagina. This report appears to be the first case of pygomelia in pigs with concurrent hermaphroditism.

The common causes of genetic abnormality in pigs include: folic acid deficiency, exposure to radiation and chemicals, toxins such as mycotoxin and viral diseases. In only 13% of congenital defects in pigs, are the causes known or believed to be heritable, or a known environmental or teratogenic agent identified [11]. The exact cause of the anomaly in this piglet is unknown. The history of the stock did not contain any prior congenital anomalies and also precluded the possibility of exposure to poisonous plants, radiation or chemicals. Although there was an outbreak of African swine fever on the farm, the farm had been rested for five years prior to re-stocking. Also, the present stocks were reported to be obtained from a new source.

Intersexuality is a frequently occurring defect in pigs with reported frequencies ranging from 0.1 percent to 1.4 percent [3]. In general, intersexes may be either true hermaphrodites, which have gonads of both sexes, or pseudohermaphrodites having either female or male gonads. The greater majority of pig intersexes have been reported to belong to be pseudohermaphrodites and have been shown to possess the normal female XX chromosome constitution. This piglet can be regarded as a true hermaphrodite owing to the presence of: a penis, inguinally located testes, bulbo-urethral gland, a pair of ovaries and a well-formed uterus with an externally located vagina. The probable cause of the intersex in the piglet may be a recessive autosomal gene with sex-limited expression either with polygenic inheritance or more likely due to recessive genes at very few autosomal loci.

The most common abnormality in the male reproductive system is cryptorchidism. It is hypothesized to be due to complete penetrant recessive genes at two autosomal loci [1]. Most cases of cryptorchidism in piglets are also associated with other abnormalities. Although the piglet can be regarded as a true hermaphrodite, the presence of inguinally located testes is suggestive that the genetic cause for the intersex might also be responsible for this abnormality.

The inheritance of polydactylism is much less clear, owing to the irregular expression of the defect. A case of autosomal dominant complete polydactyly has been reported in Papua New Guinea village pigs [5]. Although cytogenetic analysis was not done for this piglet, it is logical to presume that the skeletal anomaly was inherited and the mode of inheritance was probably complex and related to the factors responsible for the intersex in the piglet.

Finally, the presence of a unilateral renal dysplasia might have compromised the urinary functions of the piglet which probably led to the hypertrophy of the second kidney. This might explain why the piglet did not recover from a single injection of ketamine hydrochloride since the anaesthetic agent is excreted through the kidney unchanged. The vesicular structure in the abdomen will appear to be a rudimentary accessory urogenital tract or an accessory rudimentary bladder because of the predominant transitional epithelium of its mucosa later seen in the histology.

In conclusion, this report presents probably the first case of a complex malformation of true hermaphroditism and pygmelia in a pure bred Large White piglet.

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