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Case Report

Imperforate anus in a male Friesian-Holstein calf: Case report and review of literature

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A case of imperforate anus in a 3-day old male Friesian-Holstein calf was presented and treated in Bugesera district, Rwanda. The condition has rarely been reported in Africa. A brief review of the etiology, epidemiology, diagnosis, surgical and post-surgical management and classification of *anorectal malformation*s (ARM) as they occur in man and domestic animals with emphasis on imperforate anus is hereby presented. We focus on the usage of the terms imperforate anus, atresia ani and make a distinction between these terms and other ARMs. With emphasis placed on the nomenclature of ARMs and the distinction between usage of the terms *imperforate anus* and *atresia ani*, we propose that the two terms be viewed as different and standard scientific nomenclature of Type I –IV ARM be used consistently to avoid confusion. If the terms *atresia ani* and *imperforate anus* should be used, they must be used correctly and consistently.

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Introduction

Congenital malformations of the rectum and anus are referred to as *anorectal* malformations (ARM) (Beudeker *et al*., 2013; Herman & Teitelbaum, 2012). The malformations of the *anorectal* region are quite common in man and domestic animals. Earliest reports of these malformations can be seen in the works of Hippocrates (460-370 BC) and later, Aristotle in the early 3rd century BC (Tsoucalas *et al*., 2012). These malformations comprise a wide spectrum of anomalies of the *anorectum*, *urogenital* system, sacral spine and *perineal* musculature (Bhatnagar, 2015). Malformations of the rectum and anus result from disturbances in the development of the dorsal part of the hindgut (Noden & Lahunta, 1985; Hyttel *et al*., 2010). More precisely, defects in formation or shaping of *cloacal* membrane formation and subsequent breakdown into urogenital and anal openings are responsible for the numerous abnormalities of the *anorectum* (Bhatnagar, 2015).

Although rare, *ARMs* have been reported in many parts of the world including Africa. However, *ARMs*, and more specifically, *imperforate anus* has not been previously reported in Rwanda. Herein, we report a case of *imperforate anus* in a male calf belonging to

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Rwandan communal farmer that was successfully managed under field conditions.

Case presentation

We report a case of imperforate anus or Type II ARM in a male Friesian calf. The mother of the calf was a high producing cow in its 5th parity and had previously given birth to 2 males and 2 female calves which did not show any signs of *imperforate anus*. The cow had, during the first trimester of pregnancy, been rectally palpated by an inexperienced para-veterinarian for pregnancy diagnosis. The history of the bull could not be traced as the records of artificial insemination were not in order. The owner reported that she had observed a dimple in the *perineal* region which became a protrusion when the calf was straining or when pressure was applied to the abdomen. She had also noticed that the calf had no anal opening and was thus not defecating 2 days after birth.

A 3-day old male Friesian calf was presented to an ambulatory clinic veterinarian with a complaint of non-passage of feces, tenesmus, slight abdominal distention, abdominal discomfort, absence of the anus and a slight ballooning in the place where the anus was supposed to be (see plate 1 & 2).

On clinical examination, the calf was dull with shallow breaths, occasionally straining and the hair coat was rough. Palpation of the perineum revealed an anal reflex with a slight balloon emanating a sharp ping sound when tapped with a finger. A tentative diagnosis of imperforate anus was made and the calf was immediately scheduled for surgery.

The calf was sedated heavily by intramuscular injection of 0.2 mg/kg xylazine. Five minutes later, the calf was restrained by veterinary assistants and 0.2 ml/kg of lignocaine solution was infiltrated in a ring around the place where the anus was supposed to be located. The perineum was shaved with a razor blade and cleaned with gauze dipped in Povidone iodine, and sprayed with 70% isopropyl alcohol. A 15-gauge needle inserted through the center of the ballooned area in the position where the anus was supposed to be located resulted in escape of gas from the anus. After removal of the gas 2 cruciate incisions were made through the whole depth of the skin according to a method modified after Glenn and Mcsbemry (1966). The same method was also used by Suthar *et al.* (2010). Meconium was ejected violently from the anus (see plate 3). After cleaning the area, the flaps of skin were dissected out to leave a circular hole of about 1.5 cm in diameter. Three clamp forceps were inserted into the hole (see plate 3) and the epithelium of the anus held together and then simple continuous suture pattern with non-absorbable nylon was used to suture the epithelium of the inside of the anus and skin of the anus together around a small pipe.

After the surgical procedure, the operated area was cleaned and the wound was generously dusted with eye and wound powder containing oxytetracycline. Finally, methylated spirit was sprayed onto the area and mineral oil applied to the inside of the anus to aid defecation. Since the calf was still exclusively on milk, no softening of feces was necessary. The calf was released to its mother.

The wound was dressed daily with povidone iodine and an injection of 1.5 ml penicillin-streptomycin for 3 days and 1.5 ml phenylbutazone for 2 days. The sutures were removed after 2 weeks and the wound healed without complications or fecal incontinence. The calf was defecating normally after 3 weeks.

Plate 1: Back (anal) end of the calf showing a small dimple where the anus should be located

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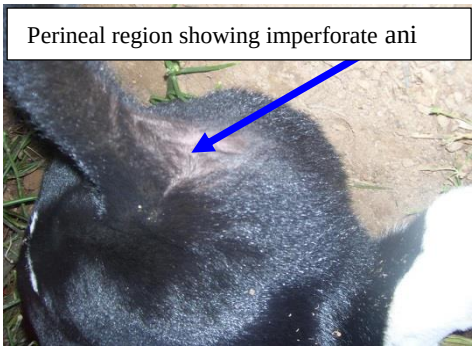

Plate 2: The imperforate anus

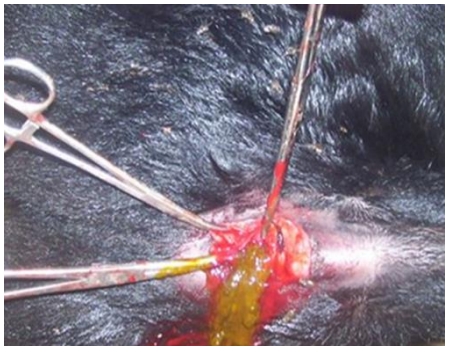

Plate 3: Opened imperforate anus perforated during the surgical procedure

Discussion

Definition

Imperafote anus is a type II ARM in which the embryonic anal membrane of a *neonate* has not yet broken down at the time of its birth.

Etiology

The etiology of this case of *imperforate anus* cannot be determined with accuracy. According to literature, the etiology of ARM is not clearly understood but it is believed to be multifactorial (Falcone *et al.*, 2007; Lombardero & Yllera, 2014; Gangopadhyay & Pandey, 2015; Wang *et al*., 2015). Workers in human and veterinary medicine (Ghanem *et al.*, 2004; Noorbakhsh, 2012; Binanti *et al.*, 2013; Wang *et al*., 2015) have fingered genetic and environmental factors, or the interactions of these factors as the major etiological risk factors. Common among these causative risk factors were maternal conditions such as obesity (Stothard *et al*., 2009), diabetes, and fever during the first trimester of pregnancy, *perineal* injuries, asthma, epilepsy, vitamin A deficiency, folic acid deficiency etc.

Viral infections have also been alleged to be responsible for malformations (Noh *et al*., 2003). According to Brenner and Orgad (2003) and Constable and Morin (2000), amniotic palpation in early pregnancy has the potential to cause ARM as a result of disruption of blood supply to the hindgut of the developing embryo.

Falcone *et al.* (2007) also pointed out genetic mutations and chromosomal abnormalities as some of the main causes of this condition. Furthermore, empirical evidence suggests a more sinister association of ARM with certain syndromes such as Townes-Broks syndrome, Currarino's syndrome (Gangopadhyay & Pandey, 2015), and Down's syndrome (Wang *et al*., 2015). Noh *et al*. (2003) pointed out that defects of anorectal and other systems may occur when a mutation occurs in a single chromosome carrying genetic information important to several metabolic pathways. In fact specific genes have been linked to specific ARM defects in mice (Herman & Teitelbaum, 2012). Given that this particular cow was pregnancy diagnosed earlier by an inexperienced para-veterinarian, we can only suspect that this pregnancy diagnosis procedure by an inexperienced para-veterinary practitioner could have been the cause of this condition

Epidemiology

Imperforate anus usually affect the terminal portion of the *cloaca*. According to literature, *ARM*s can occur anywhere from as high up as the terminal portion of the *hindgut* to the *perineum*. The higher up the digestive system the lesion is, the more complex the management required to correct it, and vice versa (Gangopadhyay and Pandey, 2015; Tsioli *et al*., 2009).

*ARM*s are quite common in cattle (Radostits *et al*, 2014), pigs (Lambrecht & Lierse, 1987) and less common in dogs and cats (Papazoglou & Ellison, 2012). It is noteworthy that Papazoglou and Ellison (2012) and Wamaitha *et al.* (2015) were in agreement that ARM are most common in pigs than other domestic animals. The condition is believed to be more common in men than in women (Herman & Teitelbaum, 2012) and in both male and female cattle (Hossain *et al*., 2014). A familial disposition has frequently been fingered in man (Herman & Teitelbaum, 2012), dogs (Papazoglou & Ellison, 2012) and cattle, particularly, West highland breed (Daradka, 2013) and Friesian-Holstein (Constable & Morin, 2000; Lombardero & Yllera, 2014). In dogs, Papazoglou & Ellison (2012) reported a higher prevalence in males than females and gave a female to male ratio of 1.79:1.

ARMs can occur as single malformations or in combination with each other or in association with other malformations such as cardiovascular (Rosenbaum *et al*, 2014), spinal-vertebral, digestive, urogenital (Binanti *et al.*, 2013; Tirtayasa *et al*., 2013) as well as gynecological malformations (Gangopadhyay & Pandey, 2015). It is therefore not surprising that this particular case is being reported in a male calf of the Friesian breed. In addition, our observations revealed that this particular case did not seem to be associated with any other systems nor was it part of any syndrome described by previous authors.

The fact that this condition was diagnosed in a Friesian male calf is not surprising as other workers (Constable & Morin, 2000; Lombardero & Yllera, 2014) have pointed out that there is a genetic predisposition in the Holstein-Friesian than other breeds of cattle. The occurrence of the condition in male calves has also been reported by other workers (Ghanem *et al*., 2004; Hossain *et al*., 2014; Suthar *et al*., 2010).

What is surprising, however, is that it has never been reported in Rwanda. Furthermore, in the authors' combined 35 years of experience, this was the first encounter of a calf with imperforate anus.

Pathogenesis

According to Noden and Lahunta (1985) the *anus* and *rectum* are formed in the following manner; the cavity at the distal end of the *hindgut* which forms the rectum and cranial part of the anus (actually the greater part of the anal canal), initially fills with epithelial cells. At an appropriate time these epithelial cells undergo apoptosis to re-establish the lumen of the rectum and cranial part of the anus. When fewer than these epithelial cells degenerate, the lumen is smaller than normal and hence atresia recti and/or anal stenosis (type I *ARM*) occurs. Atresia is a general term that describes congenital occlusion or discontinuity of the lumen of the digestive tract (Daradka, 2013). On the other hand, imperforate anus or type II *ARM* occurs when the anal membrane, a derivative of the *cloacal* membrane fails to break down before birth (McGaedy *et al*., 2006; Abouelnasr *et al.*, 2012; Acer *et al.*, 2013; Wamaitha *et al.*, 2015). Imperforate anus is the simplest of the ARM to manage. It is also the most common of the ARMs.

Diagnosis

According to literature, *imperforate ani* diagnosed by history, physical examination (Papazoglou & Ellison, 2012; Salari Sedigh *et al*., 2010), plain radiography (Tsioli *et al*., 2009), contrast radiography (Noorbakhsh, 2012; Jardel *et al*., 2013), ultrasound and magnetic resonance imaging and even computed tomography (Bhatnagar, 2015; El Mhabrech *et al*., 2015; Gangopadhyay & Pandey, 2015). Our case was diagnosed through history and physical examination alone.

Case Management and Prevention/Control

The only method of treatment for most ARMs including imperforate anus is surgery (Papazoglou & Ellison, 2012; Salari Sedigh *et al*., 2010) but requires timely and accurate diagnosis (Beudeker *et al*., 2013). The treatment may be as simple as a cruciate stab into anal location of the perineum to high-tech complicated reconstructive surgery done in stages (Nelson *et al*., 2015; Oehme, 1988). In uncomplicated cases of imperforate anus, surgery is simple, economic, and lifesaving (Daradka, 2013). However, complications leading to post-operative bowel dysfunction including fecal incontinence are not uncommon (Daradka, 2013). Other complications include wound dehiscence/infection and anal stenosis through excessive scar tissue, *megacolon* and ascending urinary tract infection (Jardel *et al*., 2013).

An antibiotic cover and non-steroidal anti-inflammatory drugs are usually administered during and after surgery (Daradka, 2013; Wamaitha *et al*., 2015).Although no specific genetic etiological cause could be definitely established in our case, we recommended that the calf be castrated as the possibility of a genetic origin for this case was still

there. As Constable and Morin (2000) and Brenner and Orgad (2003) pointed out, early pregnancy diagnosis could give rise to ARM; we also recommended to our farmer that pregnancy diagnosis be delayed to after 2 months (after organogenesis). The characteristic dimple at the expected anal location described by Hossain *et al.* (2014) gave us confidence in our diagnosis of imperforate anus. Our timely intervention was critical as the condition is regarded as fatal in males whereas fistulae connecting the anus to the vulva usually develop in females to allow for voiding of feces (Papazoglou & Ellison, 2012). The etiology of this particular condition is not clear as the mother of the calf was a pentaparous cow which had produced 4 healthy calves. However, the genetic connection could arise from the sire's side although the bull's history could not be established. The fact that the mother had produced 4 other healthy calves does not negate the possibility of the genetic defect arising from the mother since the condition is rare.

Though our surgery was performed under less than ideal conditions (no radiograph or hospital facilities) but it was both lifesaving and successful. If surgery had not been attempted on time the calf could have died within 2 weeks (Constable & Morin, 2000; Cohen & Windsor, 2013). Our success could be attributed to the fact that our aseptic procedure was strict. In addition, our case was the uncomplicated type and not accompanied by other urogenital malformations described by Ghanem *et al.* (2004). Our surgical site healed successfully without complications such as abscesses and wound dehiscence, described by others (Jardel *et al*., 2013).

We believe that the anal muscles were well developed (as shown by well-developed anal reflex) and the only problem was actually the failure of anal membrane degeneration similar to the case described by Suthar *et al.* (2010). Furthermore, our diagnosis and intervention were both timely and accurate.

Classification of ARM

Malformations of the *anorectum* in domestic animals have been typically been classified into four classes. These classes include; congenital anal stenosis (Type I); imperforate anus alone (Type II) or combined with more cranial termination of the rectum as a blind pouch (Type III); and discontinuity of the proximal rectum with normal anal and terminal rectal development (Type IV) (Binanti et al., 2013; Papazoglou & Ellison, 2012; Vallefuoco et al., 2013; Vianna & Tobias, 2005).

The terms *atresia ani, imperforate ani* and *ARM and proposed way forward*.

There is a great deal of laxity around the use of the terms *atresia ani*, *imperforate anus* and even *ARM* in human and veterinary medical literature (Binanti et al., 2013; Lombardero & Yllera, 2014; Vallefuoco et al., 2013; Vianna & Tobias, 2005). *Atresia ani* is actually the same as type I *ARM* or anal *stenosis* (Noden & Lahunta, 1985). However, many authors (Kumar et al., 2009; Papazoglou & Ellison, 2012) do not actually make the distinction between *atresia ani* and *imperforate ani* (type II *ARM*) and use the terms interchangeably. Papazoglou and Ellison (2012) have stated that failure of the breakdown of the anal membrane leads to *atresia ani* (type I ARM). This is simply incorrect if we stick to the definitions given by other authors (Binanti et al., 2013; Vallefuoco et al., 2013; Vianna & Tobias, 2005) and even his own report (Papazoglou & Ellison, 2012). Occlusion or stenosis of the terminal part of the anus cranial to a patent anal membrane results in *atresia ani* or type I ARM, while persistence (failure of breakdown) of embryonic anal membrane gives rise to imperforate anus or type II ARM). In spite of the clear classification of ARM adopted by other workers (Binanti et al., 2013; Vallefuoco et al., 2013; Vianna & Tobias, 2005), Kim et al. (2013) used the term *atresia ani* for a type III *ARM* and this loose use of terminology should be avoided.

This loose use of terminology of *ARM*s in domestic animals is bound to bring about attendant confusion. *Atresia* is a scientific general term used to describe congenital occlusion or discontinuity of the lumen of a tubular organ resulting from reduced canalization of the lumen after epithelial proliferation (Daradka, 2013). We therefore propose that the use of the term *atresia ani* be restricted to type I *ARM* in which *atresia* actually occurs.

On the other hand, we propose that the term *imperforate anus* be distinguished from *atresia ani*. *Imperforate anus* stands for the defect resulting from persistence of the embryonic anal membrane. Using the arguments advanced above and in agreement with McGaedy *et al*. (2006) and Acer *et al.* (2013), our case report was therefore one of ARM type II or *imperforate anus*.

For the rest of the ARM, we recommend the use of the terms, Type III and type IV ARM, in accordance with scientific nomenclature similar to the one described in humans by Stephens and Smith (1986) and adopted

by other workers (Binanti *et al*., 2013; Fossum, 2013; Papazoglou & Ellison, 2012; Slatter, 2003; Vallefuoco *et al*., 2013; Vianna & Tobias, 2005). Imperforate anus is common in pigs and cattle and less common in small animals.

Conflicts of interest

The authors declare that they have no financial or personal relationships which may have inappropriately influenced them in writing this article.

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