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CASE REPORT

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Surgical correction of polymelia in the perineal region of a 2-day-old indigenous bovine calf: a case report from Bangladesh

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Abstract

Background Polymelia is an occasional and congenital malformation in bovine calves, characterized by the presence of single or multiple accessory limb(s) at various body regions, which are often called the supernumerary limb(s). One of these defects is known as pygomelia, where the additional limb is attached to the pelvis within the perineal region. This study describes such a case along with surgical management.

Case presentation A 2-day-old indigenous bovine female calf of 22.7 kg BW was presented with an additional and non-functional fifth limb at the perineal region. The ectopic fifth limb was smaller in size than the other four normal limbs and was in a flexible hanging position. Other than this, the animal was apparently normal with a short and contracted tail. Clinical examinations involving the extension and flexion of the abnormal limb revealed no deep bony attachment/articulation with the pelvic girdle or the caudal spine. The curled tail having an S-shaped base indicated the skeletal defects in the caudal spine. Electrosurgery was performed under deep sedation with regional as well as local anesthesia, which exposed that the limb was internally seated into deep gluteal muscle layers with the attachments of fibrous connective tissues and cartilage to its proximal bony head and located partially to the left lateroventral extremity of the perineum without any direct involvement in the bony pelvis. Surgical excision was done to remove the supernumerary limb, and the wound was closed routinely. Postoperatively, the animal was provided with supportive medications for early healing and recovery. After 2 weeks of surgery, the calf completely recovered without any complications. The contracted tail and spine were left unaffected as no initial surgical maneuver was done due to the early age of the calf and would be considered for further long-term treatment options upon the growth and development with age.

Conclusions Pygomelia of bovine calves is one of the rare congenital defects that can be found along with other developmental anomalies. However, this defect can be successfully corrected by surgical approaches following intensive postoperative care and management.

Keywords Fifth-legged pygomelia, Congenital defect, Contracted tail, Electrosurgical interventions, Case report

Background

Polymelia is an exceptional and quite rare inherited disorder found in animals causing the additional manifestation of one or more supernumerary limb(s) or parts of limb(s) together with the normal ones (Neupane et al. 2017; Morath-Huss et al. 2019). This congenital defect is often termed according to the site of attachment of the accessory limb(s) to the body, i.e., attachment to the head (cephalomelia), back (notomelia), thorax (thoracomelia),

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and pelvis (pygomelia) (Denholm et al. 2011; Yun et al. 2015; Silva et al. 2021).

The actual reasons for this defect have not yet been clarified. However, the existing literature outline some causes, i.e., heterotopy and teratogenic impacts on the early embryonic development, genetic mutations (spontaneous or inherited), fragmentary development of conjoined twins (asymmetric and monozygotic), and other maternal abnormalities (Kim et al. 2001; Neupane et al. 2017; Morath-Huss et al. 2019). Polymelia can be categorized based on the twin malformation (symmetric or asymmetric) into two types: partial or total doubling of the axial skeleton and doubling or multiplication of limbs (Schönfelder et al. 2004). The supernumerary limb(s) can be found normal or in association with other disorders such as syndactyly, polydactyly, and ectrodactyly (Murondoti and Busayi 2001; Alam et al. 2007; Araujo et al. 2020). In addition, these types of limbs usually lack muscles having fixed joints (Denholm et al. 2011; Ali and Ibrahim 2018).

Polymelia has been reported in various species including domestic and crossbred cattle, sheep, pigs, birds, and even humans (Yun et al. 2015; Pourlis et al. 2019; Gapsiso et al. 2020; Salameh et al. 2020; Silva et al. 2021). Cattle breeds such as Holstein–Friesian, Brahma, Hereford, Angus, indigenous Korean, etc., have typically been reported with this birth defect (Kim et al. 2001; Beever 2013; Neupane et al. 2017). Among different types of polymelia, notomelia is found to be the most frequent case in bovine calves, whereas pygomelia is a relatively unusual case (Mistry et al. 2010).

The present study reports a typical case of pygomelia in a bovine calf from Bangladesh upon clinical observation and further includes the relevant surgical approaches to remove the aberrant accessory structure along with the postoperative management and outcome.

Case presentation

Case history and clinical findings

An indigenous bovine female calf aged just 2 days and of 22.7 kg body weight (BW) was referred to the Veterinary Teaching Hospital (VTH) of Bangladesh Agricultural University (BAU) with the complaint of an inborn growth of an extra limb within the perineal region in-between the two normal hind limbs (Fig. 1A). The feeding, defecation, and urination including other activities of the calf were reported normal by the animal owner. Clinical examination revealed that the limb was a supernumerary limb. The other four limbs (i.e., two forelimbs and two hindlimbs) were normal and functional. The additional fifth limb was smaller in overall length and size than the normal limbs. Anatomically

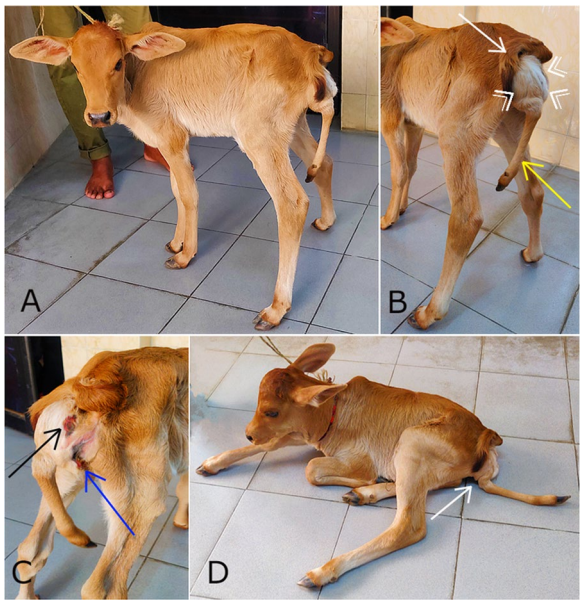

Fig. 1 The calf showing pygomelia in different positions. **A** An accessory hindlimb (small) with four normal limbs at standing position. **B** Hanging of the abnormal limb (yellow arrow) from the left lateroventral edge of the perineum (white arrow heads) below the tail (white arrow). **C** location of the limb in respect to the anus (black arrow) and vulva (blue arrow). **D** Sternal recumbency of the calf indicating the flexibility of the limb against the floor to its attachment (white arrow) within the perineal region near the caudal pelvis

it was located below the anus and close to the caudal pelvis, being left lateral to the vulva in a moveable pendulous fashion (Fig. 1B, C) and flexible to the area of attachment (Fig. 1D). Other than this defect, there was another visible skeletal malformation at the base of the tail extending up to the sacrum, which had made the tail dwarf and contracted with a characteristic S-shaped curvature at the region of the tail base and dorsal rump (Fig. 2A). Physical palpation emphasizing the extension and flexion of the limb including necessary abduction as well as adduction facilitated to detect an attachment of that limb to the left lateroventral extremity of the perineum (Fig. 2B) and aided to assume that it was not involved firmly with the internal structures, i.e., bony pelvis or caudal vertebrae. In addition, manipulation of the tail revealed quite a rigidity along both axes due to the bent stump. The clinical parameters (i.e., temperature, pulse rate, respiratory rate, and SpO_2), appetite, and hydration level of the calf were within the normal ranges. No further radiographic and hematobiochemical examinations were done, and electrosurgical extirpation of the ectopic limb was eventually considered to handle the case.

Fig. 2 Right lateral recumbency of the calf exhibiting the skeletal defect in the tail and exposing other collateral structures. **A** Short and contracted tail having a distinguishable S-shaped bending at the base dorsally to the rump (white rectangle). **B** Extension of the supernumerary limb (black arrow) exposing a loose attachment to skin and underneath tissues near to the anus (white arrow) and vulva (yellow arrow) distinct from the fairly rigid tail (blue arrow)

Electrosurgical excision of the supernumerary limb for pygomelia correction

At first, the calf was kept fasting for 8 h and then stabilized with the intravenous (IV) administration of 250 ml of 5% dextrose in normal saline (0.9% NaCl). Prior to the electrosurgery, the animal was premedicated with atropine sulfate @ 0.04 mg/kg BW (Atrovet®, Techno Drugs Ltd., Narsingdi, Bangladesh) followed by deep sedation with xylazine hydrochloride (HCl) @ 0.08 mg/kg BW (Xylaxin®, Indian Immunologicals Ltd., Hyderabad, India) through the intramuscular (IM) injections. After proper sedation and restraining, caudal epidural anesthesia was performed with 2% Lidocaine HCl (Jasocaine®, Jayson Pharmaceuticals Ltd., Dhaka, Bangladesh) and local infiltration of the same analgesic (Jasocaine®) was done in a ring block fashion surrounding the attachment of the supernumerary limb to the perineum. Following adequate regional desensitization and presurgical aseptic preparations; electrosurgery was performed by a monopolar probe-like electrosurgical scalpel (Fig. 3A) of an alternating polarity-induced active cutting and coagulation electrode along with a return (ground) electrode, having overall an electrosurgical unit [Mediton® MT-400, Class I, Type CF, 40,013 Castel Maggiore (BO) Italy via C. Bonozzi]. This device was regulated to flow the current at 50 Watt (W) and 40 W for necessary cutting and coagulation, respectively. After the electro-incision in a circular pattern over the skin of the selected area encircling the base of the limb, the surgical field was exposed clearly by using Allis tissue forceps and artery forceps. Blunt dissections of the underlying subcutis, thin muscle layers, and adjacent fibrous connective tissues were done to find out the innermost attachment of the limb. This revealed that a femur-head-like bony proximal extremity was attached internally with minor cartilages (Fig. 3B) and ends of fibrous tissue inserted muscles to the gluteal region. Surgical manipulation confirmed that there was no association or articulation of this accessory limb or its part(s) with the sacrum, pelvis, and/or caudal vertebrae during the transverse tissue dissection using the in vivo cutting effect of the electro-probe, and the coagulation effect was applied to check considerable bleeding from the operated site. Finally, the ectopic limb was excised from the perineal region. A hollow muscular cavity (Fig. 3C) in the gluteal region originated due to the removal of the deep-seated bony head of the limb, and the cavity was then flushed with normal saline followed by gentle soaking and cleaning with gauze. Thereafter the fleshy cavity was closed effectively by purse string suture (Fig. 3D) using Polyglactin 910 of size 1–0 (Vicryl™, Ethicon, J & J Medical Devices Companies, USA). Muscle layers were closed in a simple continuous suture pattern (Fig. 3E) using Vicryl™ (1–0), and the skin was closed by simple interrupted sutures (Fig. 3F) using nylon thread of size 2–0 (Nylon Monofilament, MediPharm Biotech. Co. Ltd., Shanghai, China). Finally, the excised limb was discarded (Fig. 3G) in the VTH dumping pit without further organ dissection and laboratory examinations.

Postoperative care and outcome

After surgery, the calf was again provided with 250 ml of IV 5% dextrose in normal saline. A course of ceftriaxone sodium @ 15 mg/kg BW (Trizon Vet, ACME Laboratories Ltd., Dhaka, Bangladesh) for 7 days, ketoprofen @ 3.3 mg/kg BW (Ketovet, Techno Drugs Ltd., Narsingdi, Bangladesh) for 5 days, and pheniramine maleate @ 1 mg/kg BW (Antihista-Vet®, Square Pharmaceuticals Ltd., Dhaka, Bangladesh) for 7 days was maintained postoperatively through IM injections. Fly repellent was regularly used in the cattle shed to prevent myiasis, and 5% povidone-iodine (Viodin® 5% Ointment, Square Pharmaceuticals Ltd., Dhaka, Bangladesh) was topically used twice daily for 7 days. After 10 days of surgery, a combined preparation of vitamin A, D_3, and E was provided orally to the calf twice daily for the next 15 days. On the 14th postoperative day, the external sutures were removed, and the animal seemed to be normal with postures and gestures. One month following surgery, the

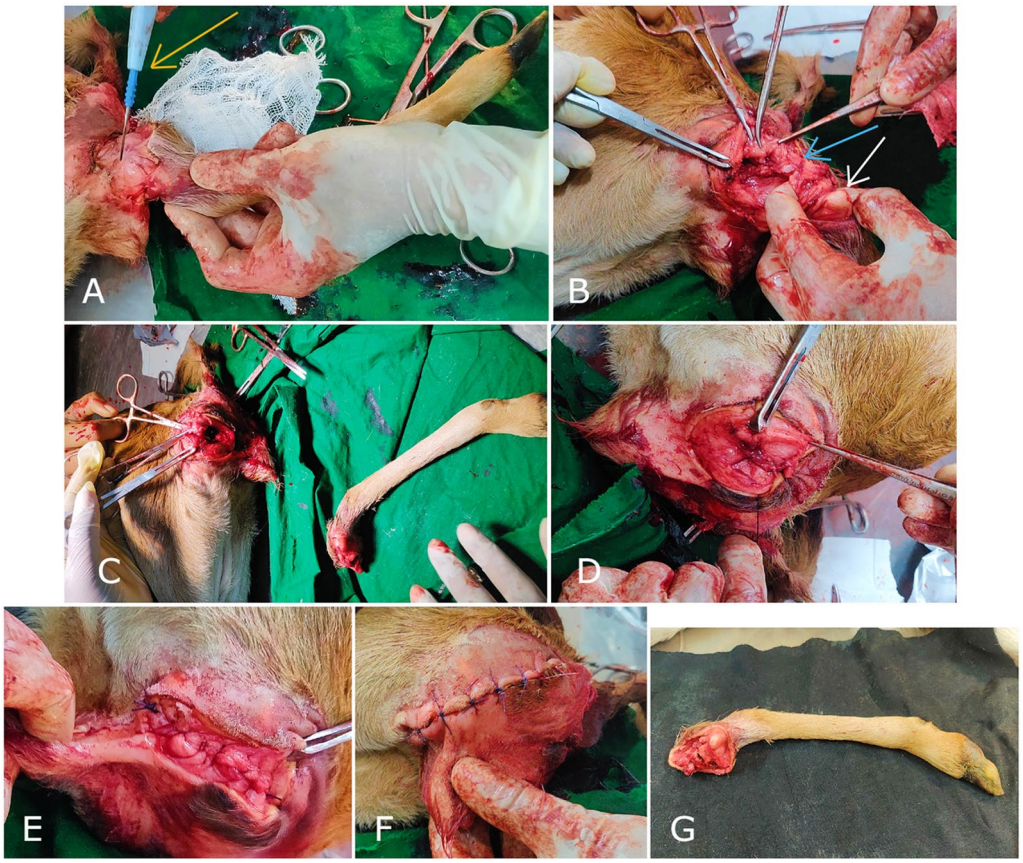

Fig. 3 Different phases of electrosurgery to excise the fifth additional limb along with the final wound closure. **A** Monopolar electro-probe (yellow arrow) having simultaneous cutting and coagulation capacities applied locally for the transection of the limb. **B** Exposure of the femur-head like bony prominence (white arrow) detached from thin cartilages (blue arrow) and fibrous connective tissues adjacent to the structure. **C** A muscular hollow in the gluteal region derived from the surgical removal of the limb. **D** Closure of the hollow cavity by purse string suture. **E** Closure of the surrounding muscles by simple continuous suture. **F** Skin closure by simple interrupted sutures. **G** The discarded limb

owner reported no further complication except the prior congenital defect in the tail base and sacrum, which had been left untreated at the time of pygomelia surgery, and it would be considered further with priority based on sufficient maturity and suitable age for the spine and skeletal developments.

Discussion

The congenital abnormalities are much more frequent in domestic ruminants and have been reported to be about 2% to 3.5% of all births (Muirhead et al. 2014). Among those, anomalies involved in the limbs and musculoskeletal systems (particularly the axial and appendicular skeleton) of newborns are common in bovine species (Kim et al. 2001; Alam et al. 2007; Abdel-Hakiem and Elrashidy 2017). These defects are usually responsible for economic losses to the food-animal farms due to the reduced market values (Neupane 2018), incapability of production and performance, stunted growth, etc., of the affected animals. For these reasons, congenitally defective animals are often euthanized/culled or slaughtered after a short-term fattening (Morath-Huss et al. 2019). In contrast, genetically exceptional animals having the extra growth of limb(s), head, horn, eye, or other organs are supposed to be the worship materials for wish fulfillment by some tribal and regional people (Green 2021); however, science suggests these as superstitions.

Polymelia being an inheritable defect with the additional limb(s) in animals, especially in cattle, has been found in various crossbred and exotic groups and is not a fatal defect. In addition, among the indigenous species; only the Korean indigenous cattle have been reported with this defect (Kim et al. 2001; Yun et al. 2015). In this study, an indigenous Bangladeshi female bovine calf was found to have a special type of polymelia called pygomelia showing a non-functional and smaller fifth limb, and to the best of our knowledge, this is the first observation of such a case in the native bovine species in Bangladesh.

Occasionally polymelia is combined with other skeletal disorders, such as the absence of certain bone(s),

or underdeveloped/maldeveloped bones or bony fragments (Murondoti and Busayi 2001; Shojaei et al. 2007; Montalvo et al. 2014), and/or with different genetic disorders (Ajadi and Olaniyi 2018; Daneze and Brasil 2018; Liu et al. 2019). In addition, the affected animals may have simultaneous incidences of ectopic lungs, ectopia cordis, atresia ani, and rectovaginal fistula (Yun et al. 2015; Ali and Ibrahim 2018). In the present study, the pygomelia in the calf was associated with another skeletal defect involved in the caudal spine and sacrum, which had affected the tail to become somewhat contracted and short in length.

As a certain type of polymelia, this case (pygomelia) represented the extra growth of an entire but shorter hind limb in the perineal region between the normal hindlimbs of the calf, being correlated with the findings of other researchers (Mistry et al. 2010; Ajadi and Olaniyi 2018; Singh and Vikram et al. 2022). Apart from pygomelia, there are frequent reports of other types of polymelia. Notomelia is the attachment of the additional limb(s) to the region of the embryonic notochord that helps to develop the axial skeleton and intervertebral disk(s); cephalomelia represents an attachment of the limb(s) to the head between the horn/horn-buds or caudally to the skull, and thoracomelia means this type of limb attachment to one or both lateral/medial side(s) of the thorax (Sinowatz 2010; Muirhead et al. 2014). In the case of symmetrical or asymmetrical conjoined twins mediated polymelia in cattle, the additional growth of six or eight limbs is found, whereas only one or two supernumerary limb(s) can be observed in heterotopic polymelia (Yun et al. 2015). These defects are often due to some sex-linked recessive traits (Mistry et al. 2010) and are usually diversified with animal species, breed, geographical distribution, and other environmental factors (Muirhead et al. 2014).

The pygomelia mentioned in this case might be the consequence of the presence of certain recessive genes either from the dam or sire ancestors, or due to some embryonic and teratogenic factors during the maternal period from exogenous or endogenous origins. Previously, genetic analysis for polymelia in Angus cattle showed evidence of a developmental duplication (inherited mutation) in the novel *NHLRC2* gene responsible for this defect, which was found to correlate with those of Holstein calves experimented to determine the genetic loci of polymelia (Neupane et al. 2017). Although our present study did not focus on the genetic characterization of pygomelia in that native Bangladeshi calf, this might be the further interest of this study. Determination of the genetic basis of pygomelia in indigenous Bangladeshi cattle would be helpful for selective breeding purposes by identifying the carriers to avoid these hereditary disorders.

During the clinical examination, the diagonal rotation of the accessory limb including selective extension, flexion, abduction, and adduction was carried out to manually estimate the degree of its attachment, root of articulation and stump, and internal tissue involvements to the perineum near the caudal extremity of the bony pelvis. This study did not involve any radiographic examination for pygomelia diagnosis due to the very early age (only 2-day-old) of the calf. In fact, the younger animals are much more sensitive to the radiation of X-rays, and this might cause harmful effects on the immature as well as growing cells of the neonates' bodies. However, ultrasonography or safe CT scanning can be performed in this context, although we did not use these in our study. Hematobiochemical examinations were not performed as the animal was found apparently normal with preferable clinical parameters.

Electrosurgery was performed to reduce the hemorrhage and to ensure minimum tissue damage in a least invasive way, and the exchangeable cutting and coagulation effects using the mentioned electric energy are consistent with other studies (Alkatout et al. 2012; Watts 2018; Thakare et al. 2022). As far as we know, this type of electrosurgical intervention for pygomelia correction has not yet been reported.

During the surgery, blunt and transverse dissection aided to detect thoroughly the attachment of the limb to the perineal region that revealed an association with thin cartilages, fibrous tissues, and muscles free from any involvement to the joints or bones of the pelvis, sacrum, and caudal part of the vertebral column. However, we did not perform any anatomical dissection, imaging, or laboratory examinations of the severed limb due to the unwillingness of the owner.

This case revealed an abnormality in the skeletal development of the caudal vertebrae and part of the sacrum presenting an S-shaped bending of the tail having a notable rigidity and squat appearance. This might be attributed to some anomalies during the early embryonic development within the mesodermal derivatives. Although the aforementioned ectopic limb was surgically excised, this curled tail was kept untreated at the time of the study because there might be further chances to decide the fate of the tail based on the body stance, locomotory balance, and musculoskeletal developments with age. Reconstructive surgery or docking or leaving it intact for the rest of its life would be the possible option(s) accordingly.

It might be expected that there would be no long-term major complications or obstacles regarding the reproductive behavior and performance of the calf after maturity

as there was no deeper involvement of the excised aberrant limb to the bony pelvis and pelvic outlet (birth canal) or caudal vertebra(e) or other visceral genital organs. In addition, as there was no neuromuscular dysfunction in the pelvic and perineal regions following surgery; the act of parturition in the future might not be affected as well. Besides these, from a negative perspective, there might be the chances of difficulties in matting and thereafter dystocia if there are considerable defects further seen in the pelvic canal unpredictably including the internal and external genitalia due to congenital or other abnormalities. Moreover, the contracted tail might be a definite concern if it carries the same defect and rigidity with a larger size after growth and development. Because the tail should be lifted to expose the external genitalia, i.e., the vulva and vagina during natural matting or artificial insemination, and again during parturition. That time, docking of the tail can be considered to overcome this trouble.

The presurgical administrations of atropine sulfate and xylazine HCl for premedication and sedation, and 2% lidocaine HCl for caudal epidural anesthesia and ring block for the perineal surgery are in agreement with those protocols of different research (Ismail 2016; Munif et al. 2022a). The used suture materials and suture patterns, in this case, are consistent with other reports from various experiments (Ali and Ibrahim 2018; Ryu et al. 2018; Munif et al. 2022b).

Postoperatively, there might be the chances of infections, inflammation, and myiasis. To prevent these coincidences and also to facilitate early healing and recovery, the animal was treated with supportive medications along with maintaining proper hygiene of the animal shed. Besides these, fluid therapies were provided during the presurgical and postsurgical periods to check dehydration and balance fluid, electrolytes, and energy to prevent perioperative incidences such as hypovolemia and shock. Vitamin supplementations were given to promote the growth and development of the animal.

Conclusions

Pygomelia is a rarely found case of congenital malformations in indigenous Bangladeshi cattle at field and farm levels. It might be associated with other musculoskeletal defects. Electrosurgery can be an effective method for the clinical correction of pygomelia. Complete recovery following surgery can be achieved by ensuring proper postoperative medications and intensive management.

Abbreviations

BAU	Bangladesh Agricultural University
BW	Body weight
Co.	Company
CT	Computed tomography
HCl	Hydrochloride
IM	Intramuscular
IV	Intravenous
Ltd.	Limited
NaCl	Sodium chloride
NHLRC2	*NHL* Repeat containing 2
SpO_2	Saturation of peripheral oxygen
VTH	Veterinary Teaching Hospital
W	Watt

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Author contributions

MRM performed the clinical examinations and diagnosis, anesthetic and surgical procedures, manuscript writing, and finalization. MSS and AH assisted during the surgery and postoperative follow-up, document collection, and contributed to the literature review. All authors read and approved the final manuscript.

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Availability of data and materials

All the data and supporting files have been presented within the case report.

Declarations

Ethics approval and consent to participate

This work was approved by the authority of the VTH of BAU, and consents for participation were obtained from the concerned persons.

Consent for publication

Informed consent for publication was obtained from the animal owner.

Competing interests

The authors declare that they have no competing interests.

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