Case Report

CO_2 laser clitorectomy for clitoral hyperplasia containing os clitoris and vulvoplasty for recessed vulva in a female pseudohermaphrodite dog

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Abstract

A 1-year-old spayed female Welsh corgi presented with a history of a protruding enlarged clitoris, frequent licking of the vulvar region, pain on urination, and mucopurulent vulvar discharge. The dog was diagnosed with clitoral hypertrophy with os clitoris and a recessed vulva with excessive perivulvar skin. Additionally, female pseudohermaphroditism was confirmed through physical and histopathological examinations, karyotyping, and sex-determining region Y (SRY) gene-specific polymerase chain reaction (PCR). To alleviate the clinical signs, the dog underwent clitorectomy using CO_2 laser and vulvoplasty. Clitorectomy performed using CO_2 laser allowed reduction of postoperative pain, swelling, and intraoperative hemorrhage. At 3 postoperative days, there was a significant improvement in the patient's clinical signs. This report suggests that the CO_2 laser can facilitate correction of clitoral abnormalities compared with amputation surgery using a scalpel and monopolar electrocautery.

Keywords: clitoral hypertrophy, CO_2 laser, female pseudohermaphroditism, os clitoris, recessed vulva

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Introduction

Pseudohermaphroditism is defined as a disagreement between gonadal and phenotypic sex. Accordingly, female pseudohermaphroditism (FPH) involves ovaries as gonads and masculinized genitalia. Clitoral hypertrophy with os clitoris in dogs is typically considered a disorder of sex development (DSD) or a masculinized constitution (Scarlett *et al*., 2018; Margot and Romain, 2012; Daniel and Hannah, 2014). Theoretically, masculinization of the phenotype in FPH is known to result from *in utero* exposure to androgen sources (Scarlett *et al*., 2018; Margot and Romain, 2012). The sources of androgen include exogenous exposure or adrenal hyperplasia (Scarlett *et al*., 2018; Margot and Romain, 2012; Daniel and Hannah, 2014). Exogenous exposure to androgen can result from maternal systemic or iatrogenic administration of a steroid hormone, such as progesterone or corticosteroid. Androgen overproduction in maternal or fetal adrenal disorders, such as adrenal hyperplasia or neoplasia, can result from over-secretion of the androstenedione that is converted into the androgen. (Margot and Romain, 2012).

The protrusion of hypertrophied clitoris containing os clitoris from the vulvar labia may cause chronic irritation, self-trauma, vulvar discharge and dysuria (Scarlett *et al*., 2018; Daniel and Hannah, 2014). Clitoral abnormalities that induce clinical signs and are unresponsive to medical therapy can be treated through a surgical procedure termed clitorectomy (Scarlett *et al*., 2018).

Laser surgery, especially with CO_2 laser energy, has the advantage of reducing postoperative pain (Noel and Peter, 2006). Additionally, using CO_2 laser allows adequate hemostasis with minimal thermal damage to adjacent tissues and the reduction of postoperative swelling by sealing lymphatics (Noel and Peter, 2006). Given the highly vascularized and innervated nature of the clitoris, minimal manipulation of the surgical site is necessary to avoid undesirable complications, including postoperative pain, swelling and intraoperative hemorrhage (Scarlett *et al*., 2018; Daniel and Hannah, 2014).

Previous reports have described several techniques for clitoral amputation to control intraoperative hemorrhage (Scarlett *et al*., 2018; Daniel and Hannah, 2014); however, they have several limitations. Suturing the clitoral base and oversewing amputated defects often require additional surgical procedures to secure the surgical site, including episiotomy (Scarlett *et al*., 2018). Compared with the CO_2 laser, using a scalpel for surgical resection involves intraoperative hemorrhage, postoperative pain and swelling (Jose *et al*., 2009). Moreover, the disadvantages of clitorectomy using monopolar electrocautery compared with using CO_2 laser include more thermal damage and the induction of muscle contraction by nerve stimulation, which can induce iatrogenic damage to the adjacent tissues, especially upon clitoral amputation adjacent to the urethra (Edward *et al*., 2007; Chaoyang *et al*., 2015).

A recessed vulva is defined as an excessive perivulvar skin fold that partially or completely covers the small or juvenile vulva (Barbara *et al*., 2001). This condition increases the susceptibility of the perivulvar environment to infection and inflammation, which can diminish clinical signs associated with clitoral abnormalities (Barbara *et al*., 2001; Hammel and Bjorling, 2002). A surgical option termed as vulvoplasty can successfully reduce related clinical signs (Barbara *et al*., 2001; Hammel and Bjorling, 2002; Scarlett *et al*., 2018).

This article aims to describe successful surgical correction of genital abnormalities in a female pseudohermaphrodite dog using CO_2 laser clitorectomy and vulvoplasty without postoperative complications.

Case description

A 12-month-old spayed female Welsh corgi dog presented with an enlarged clitoris, severe pain upon vulvar palpation, stranguria and perivulvar dermal ulceration (Fig. 1A). The dog was spayed early at the age of 4 months. A tubular structure lateral to the uterine horn, which was suspected to be the gubernaculum, was recognized from the ovary to the inguinal canal during the spaying surgery. Histopathological examination of the spayed ovaries revealed normal immature ovarian tissue (Fig. 2A). For 8 months after the spaying surgery, the clitoral size remained the same, with the clinical signs deteriorating over time.

On physical examination, the dog had a hypertrophied clitoris protruding from the vagina and mucopurulent discharge around the clitoris (Fig. 1B). Additionally, there were excessive perivulvar skin folds partially covering the vulvar area (Fig. 1C). The urethral opening was adjacent to the clitoris but not within the clitoris. CT examination revealed a hyperattenuating curvilinear structure in the clitoris without remnant internal genital organs (Fig. 1D). To identify chromosomal and genetic sex, karyotyping and SRY gene-targeted PCR were performed using a blood sample collected from the jugular vein. Twenty metaphases were observed in the submitted blood lymphocyte cultures; further, there was a normal female karyotype (2n = 78, XX) (Fig. 2B). For the SRY gene-targeted PCR, a pair of primers was designed to detect the canine SRY gene-coding region (GenBank accession number: AF107021) from 325bp to 779bp (5′ to 3′). It revealed that the patient was negative for SRY (Fig. 2C). Based on these chromosomal, genetic, histological, gonadal and phenotypic sex results, the patient was diagnosed with FPH.

Antibiotics and analgesics were administered for 14 days to control the clinical signs. Although there was a mild decrease in the mucopurulent discharge, stranguria and pain reaction on palpation persisted. Since there was no significant response to medical therapy, surgical treatment was chosen. Before surgery, the patient was administered with propofol (Anepol injection, Hana Pharm. Co. Ltd., Seoul, Republic of Korea) for anesthesia induction and isoflurane (Ifran, Hana Pharm. Co., Ltd.) in oxygen for anesthesia maintenance. The dog was positioned in ventral recumbency with the hind limbs placed at the end of the surgical table, followed by routine preparation of the perineal region. A pulse-string

suture was performed in the anal orifice. Further, a 6-French urethral catheter was inserted from the urethral opening to prevent iatrogenic urethral damage during surgery since the urethral opening was closed to the enlarged clitoris. Three stay sutures were placed in both the vulvar labia and clitoral tip. Sterile gauzes were packed in the vaginal opening before using the CO_2 laser (Ultra-25 Plus, Union Medical Co. Uijeongbu-Si; Republic of Korea). The vestibular plate adjacent to the urethral opening was dissected from the clitoris. The clitoral base was amputated from the vaginal mucosa using CO_2 laser (Fig. 3A and B). Subsequently, vulvoplasty was proceeded with to treat the recessed vulva. Histopathological examination of the amputated clitoris revealed lymphoplasmacytic clitoris without evidence of malignancy and a well-differentiated bone within the clitoris, which is consistent with os clitoris (Fig. 3C).

The patient recovered uneventfully after anesthesia. At 24 postoperative hours, there was a significant reduction in the pain reaction on palpation of the surgical sites without remarkable findings. The patient was discharged 3 days after surgery. There were no ambulation or urination problems. The surgical site showed mild swelling; however, there were no signs of infection or dehiscence (Fig. 4). Reexamination at one postoperative month revealed the resolution of the clinical signs. A phone interview conducted at 6 months postoperatively revealed that there were no complications.

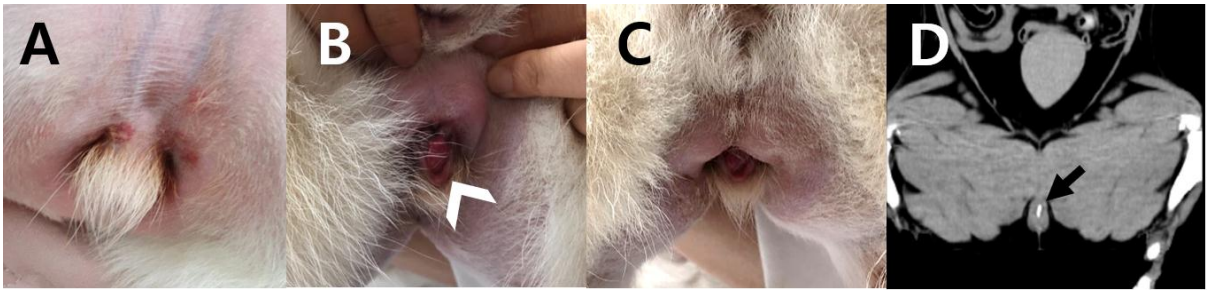

Figure 1 Preoperative examination of the perivulvar region. (A) Perivulvar dermal ulceration on the ventral perivulvar region. (B) The hypertrophied clitoris protruding from the vagina (white arrowhead). A solid material object is palpated on the ventral clitoral side. (C) Recessed vulva partially covered by excessive perivulvar skin folds. Mucopurulent discharge was stuck in the folds. (D) CT scan of the patient with clitoral hypertrophy containing os clitoris. A hyperattenuating curvilinear structure was observed in the clitoris (black arrows).

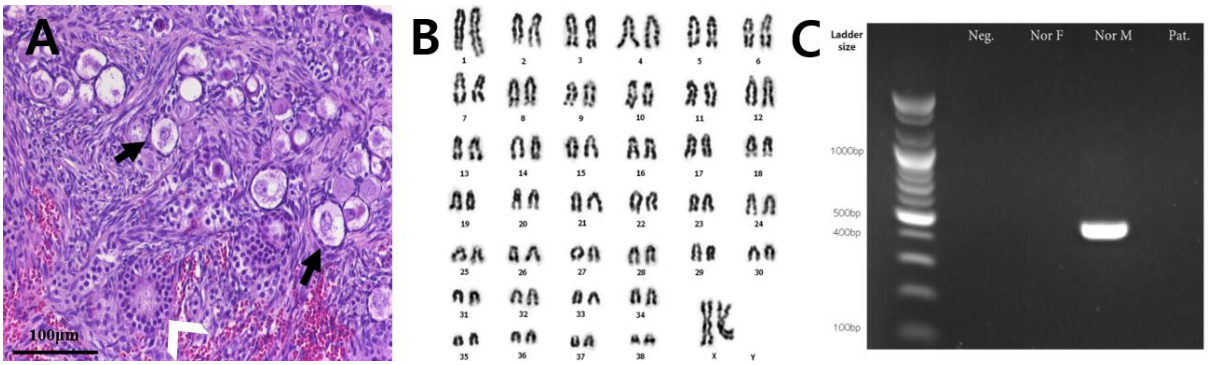

Figure 2 Genetic and histopathological examination of the patient to determine the genetic and gonadal sex. (A) Histopathological examination of the ovaries revealed normal ovarian tissues. Normal development of the primary follicle from primordial cells. Primordial cells (Black arrows). Primary follicle which contains the oocyte surrounded by granulosa cells (White arrowhead). (Scale bar = 100 μm, 200x magnification.) (B) Karyotyping results were obtained using the submitted blood lymphocyte culture (2n = 78, XX). (C) Agarose gel electrophoresis of the PCR product from SRY gene-targeted PCR. DNA extracted from peripheral blood of a normal female and male dog was used as the template for PCR (Lane 2, Nor F = normal female dog; Lane 3, Nor M = normal male dog). The PCR results were negative for the SRY gene (Lane 4, Pat. = patient). DNA free-pure distilled water was used in PCR as a negative control (Lane 1, Neg. = negative control).

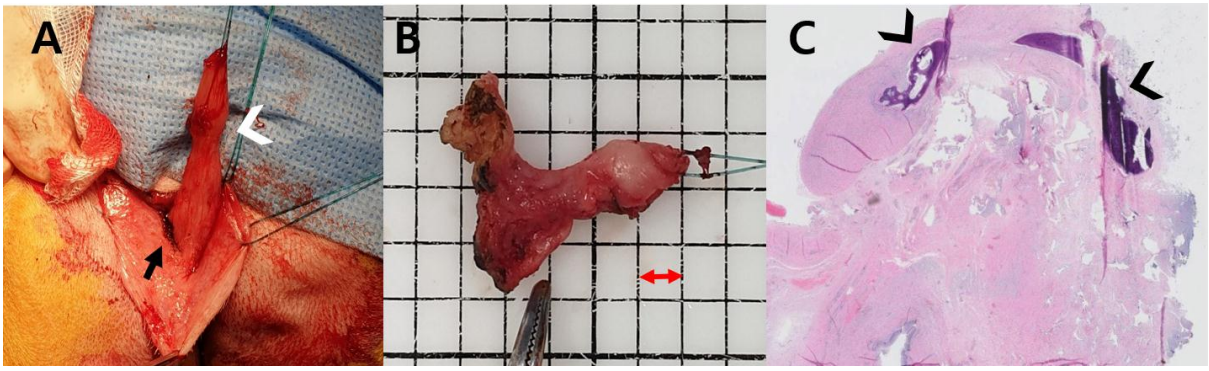

Figure 3 Surgical image of the clitorectomy and histopathological results of the amputated clitoris. (A) The clitoral base was identified by clitoral retraction and amputated from vaginal mucosa using CO_2 laser. There was little hemorrhage from the cut plane of the clitoris. Retracted hypertrophied clitoris with os clitoris (white arrowhead). The cut plane of the base of clitoris (black arrow). (B) The length of the amputated clitoris was approximately 3 cm. The squares in the background are about 0.5 cm (Red two-way arrow). (C) Histopathological examination revealed lymphoplasmacytic clitoris without evidence of malignancy and a well-differentiated bone within the clitoris, which was consistent with os clitoris (10x magnification). Bone materials within clitoris. (Black arrowheads).

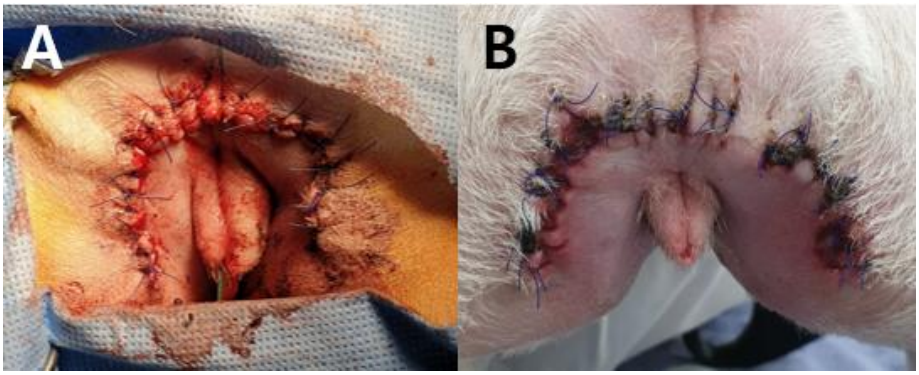

Figure 4 Postoperative monitoring of the perineal region. (A) There was remarkable swelling after clitorectomy with concurrent vulvoplasty. (B) At 3 postoperative days, the swelling was reduced and there was mild pain on palpation. There was an improvement in clinical signs related to perivulvar dermatitis and chronic UTI.

Discussion

Clitoral hypertrophy and bone tissue development in the clitoris, which is also known as os clitoris, are defined as phenotypic sexual masculinization of the clitoris (Scarlett *et al.*, 2018). In normal sexual development, chromosomal sex is determined during fertilization, which directs gonadal tissue development in either the testis or ovary. Subsequently, internal and external reproductive organs that comprise the phenotypic sex develop based on the gonadal tissue type. DSD are characterized by inconsistencies among the chromosomal, gonadal and phenotypic sex. The inconsistency between the gonadal and phenotypic sex is referred to as pseudohermaphroditism (Poth *et al.*, 2010). FPH or 78, XX DSD is characterized by ovaries with masculinized internal or external reproductive organs without chromosomal abnormalities. Reported phenotypic abnormalities in FPH vary from clitoral hypertrophy to the formation of cloaca-like structures resulting from the fusion of labioscrotal folds (Waghmare *et al.*, 2010). In our case, a series of examinations revealed external genital abnormalities and an abnormal structure along the uterine horn, which was suspected to be a gubernaculum. Based on these findings, our patient was diagnosed with FPH or 78, XX DSD, as previously described (Poth *et al.*, 2010).

The protrusion of hypertrophied clitoris from the labial cleft, especially a clitoris-containing os clitoris, is vulnerable to irritation, exposure and self-trauma (Scarlett *et al.*, 2018; Daniel and Hannah, 2014; Poth *et al.*, 2010). Histopathological examination of the amputated clitoris confirmed lymphoplasmacytic clitoris with os clitoris, which implied chronic inflammation and persistent irritation. In case of clinical signs or unresponsiveness to medical therapy, patients with hypertrophied clitoris with or without os clitoris are treated using surgical clitorectomy (Scarlett *et al.*, 2018).

The clitoris is a homolog of the male penis and contains cavernous erectile tissue lined by a subepithelial layer comprised of small vessels and capillaries for blood supply (Morio, 1955). Additionally, numerous sensory nerve endings originating from the pudendal nerve are distributed along the clitoral glans (Morio, 1955). Therefore, it is necessary to consider surgical clitoral amputation with minimal perioperative hemorrhage, postoperative swelling and pain. Accordingly, several surgical techniques have been described including root ligation before clitoral amputation, suturing the defect of the amputated clitoris using the vaginal vestibular mucosa layer and partial clitoral resection or episiotomy to secure the clitoral base, which is less vascularized (Scarlett *et al.*, 2018; Daniel and Hannah, 2014).

In human medicine, CO_2 laser is actively used for soft tissue amputation, including oral mass removal, tonsillectomy and hemorrhoidectomy (Pandini *et al.*,

2006; López-Jornet and Camacho-Alonso, 2013; Magdy *et al*., 2008). In soft tissue surgery, a 10,600-nm wavelength CO_2 laser is highly absorbed in water, which forms approximately 90% of the volume of a soft tissue cell. Since the high absorption of water allows minimal laser scattering to lateral tissues to a degree of 0.05 to 0.1 mm, the CO_2 laser is useful for controlling a beam for incision, excision and ablation of target soft tissue (Noel and Peter, 2006; Holt and Mann, 2002) Small blood vessels with a diameter of up to 0.5–0.6 mm and lymphatics are sealed by vaporization of tissues affected by CO_2 laser. This sealing effect induces intraoperative hemostasis, improves surgical site visualization and reduces postoperative swelling. The CO_2 laser prevents initiation of a sensory nerve action potential by sealing nerve endings. Moreover, the vaporization of other microorganisms, including bacteria, fungi and viruses, maintains a sterile environment during urogenital surgery (Noel and Peter, 2006).

Compared with scalpel resection, the CO_2 laser allows more rapid and simpler ablation without the need for additional hemostasis and suturing at the end of the surgery (Noel and Peter, 2006). Additionally, the sealing effect of CO_2 laser cannot be achieved in sharp amputations using a scalpel (Jose *et al*., 2009, Pandini et al., 2006). Monopolar electrocautery has also been previously used for clitoral amputations (Scarlett *et al*., 2018; Daniel and Hannah, 2014). The electrosurgery applies electric currents passing through the patient between the handpiece and the ground plate or handpiece tips (Edward *et al*., 2007). The resistance of electric currents in tissues generates heat for coagulation and cutting the target (Edward *et al*., 2007). Accordingly, compared with CO_2 laser surgery, electrosurgery generally induces more lateral thermal damage; further, muscular contracture caused by electrical stimulation of the peripheral nerve hinders precise targeting of the amputating clitoris (Edward *et al*., 2007; Chaoyang *et al*., 2015). In our case, there was a risk of urethral damage during clitoral amputation given the short distance between the enlarged clitoris and urethral opening. Using CO_2 laser allowed minimal lateral thermal damage and precise targeting of the muscular vaginal tissue. Moreover, there was a significant postoperative reduction in pain during urination. The postoperative swelling reduction contributed to the rapid improvement of clinical signs.

A recessed vulva is defined as an excessive perivulvar skin fold that partially or completely covers the small or juvenile vulva (Barbara *et al*., 2001). Excessive perivulvar skin folds can disturb urine outflow, which causes urine accumulation in the perivulvar skin fold. Frictional irritation of the skin folds combined with fluid retention contributes to the induction of perivulvar dermatitis and ulceration (Barbara *et al*., 2001, Scott and Dale, 2002) A previous study reported that the main complaints caused by recessed vulva were perivulvar dermatitis and urinary incontinence/chronic urinary tract infection (UTI) in 59% and 56% of the patients, respectively (Scott and Dale, 2002). Skin fold removal can improve clinical signs, including perivulvar dermatitis and chronic UTI (Barbara *et al*., 2001; Scott and Dale, 2002) However, in our case, persistent urine scalding and perivulvar dermatitis related to the recessed vulva complicated the management of clinical signs associated with clitoral abnormalities. Clitorectomy with vulvoplasty could effectively control persistent clinical signs in cases of concurrent clitoral abnormalities with a recessed vulva.

In conclusion, this article describes a dog with FPH that showed genital abnormalities, including clitoral hypertrophy, os clitoris and recessed vulva. The clinical signs resulting from these genital abnormalities were persistent and difficult to manage with medical treatment. Surgical treatment significantly resolved the clinical signs without postoperative complications. Compared with amputation surgery using a scalpel and monopolar electrocautery, clitorectomy using a CO_2 laser allows control of intraoperative hemorrhage, postoperative pain and swelling. This report suggests that the CO_2 laser is a useful surgical tool for correcting clitoral abnormalities.

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