Prosthetic Vaginal Dilator- A Case Report

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Citation

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Abstract

Müllerian agenesis is a congenital malformation in women. It is characterized by a failure of the Müllerian ducts to develop, which results in a missing uterus and variable malformations of the vagina. It is also the second most common cause of primary amenorrhea. This clinical report describes the fabrication of prosthesis, by a maxillofacial prosthodontist that is used in the non surgical management of Müllerian agenesis to aid in the dilation of a narrow vaginal opening.

INTRODUCTION

Müllerian agenesis is also referred to as CAUV (Congenital Absence of the Uterus and Vagina), MA (Müllerian Aplasia), GRES (Genital Renal Ear Syndrome), and MRKH (Mayer-Rokitansky-Küster-Hauser Syndrome). This syndrome is subdivided into two types: Type I (isolated) or Renal Dysplasia and Type II or MURCS association (Müllerian duct aplasia, Renal dysplasia and Cervical Somite anomalies)[1]. MRKH or Mayer-Rokitansky-Küster-Hauser Syndrome, was named after August Franz Joseph Karl Mayer, Carl Freiherr von Rokitansky, Hermann Küster, and G. A. Hauser^[2]. It is a congenital malformation in women characterized by a failure of the Müllerian ducts to develop, resulting in a missing uterus and variable malformations of the vagina. Müllerian agenesis has been considered as a sporadic anomaly^[3], but the increase in familial cases now supports the hypothesis of a genetic cause. It is transmitted as an autosomal dominant trait with incomplete penetrance and variable expressivity^[4]. The Müllerian ducts (or paramesonephric ducts) are paired ducts of the embryo (tissue of mesoderm origin) that run down the lateral sides of the urogenital ridge and terminate at the Müllerian eminence in the primitive urogenital sinus. They are present in both sexes but in the females, they will develop to form the fallopian tubes, uterus, and the upper portion of the vagina, and in males they degenerate.

It is reported to occur as a case in 4000-10,000 live female births. It is the second most common cause of primary amenorrhea^{[2] [5] [6]}. Generally patients with Müllerian agenesis present with primary amenorrhea, presenting with are 46 XX karyotype ^[7] with normal functioning ovaries and

no signs of androgen excess. [8]. In the year 1969 J.M. Tanner studied the development of female secondary sexual characteristics on the basis of height, breast development and pubic hair and classified them into 5 stages [9]. Based on this classification, these patients are categorized under Tanner stage 5 which includes completed puberty with typical secondary female sexual characteristics such as pubic hair and breast development. A normal external genitilia with a reduced vaginal opening measuring about 2-7 cm deep referred to as the vaginal dimple, is also seen. Other less common symptoms include, renal problems, loss of hearing, and skeletal malformations. Typically, the vagina is shortened. Medical examination supported by gynaecologic ultrasonography demonstrates a complete or partial absence of the cervix, uterus, and vagina [6] [10] [11]. The treatment involves the creation of a neovagina which allows the patient to perform sexual intercourse with her partner. As this syndrome can cause severe psychological stress in a young woman, it is essential for the patients and their families to attend counselling before proceeding for treatment^[1]. Family history also plays an important role in the examination process. Basile et al have stated that 30- 40 % of patients with MRKH syndrome are associated with congenital anomalies of upper urinary tract. It has also been stated in their study that 40-50-% patients were associated with renal agenesis or pelvic kidney (ectopic kidney) [12]. Hence these patients and their relatives must be investigated for abnormal renal conditions. Based on the family history the patient must undergo diagnostic tests such as urography and CT scan of the abdomen^[12]. Apart from these, there are other diagnostic methods to confirm the presence of this syndrome, some of them being transabdominal

ultrasonography, MRI, celioscopy, and the biologic status of the patient. Amongst these the 3 dimensional ultrasound may be a more sensitive diagnostic tool^[13]. The differential diagnosis of this syndrome includes the congenital absence of the uterus and vagina (aplasia or agenesis), isolated vaginal atresia, WNT4 defects, and androgen insensitivity syndrome AIS or Müllerian derivative aplasia^[14]. WNT4 is another developmental gene, belonging to the WNT family of genes that regulate cell and tissue growth and differentiation during embryogenesis.^[1].

The use of a vaginal dilator is a non surgical method of management for patients with complete Müllerian agenesis. A maxillofacial prosthodontist plays an important role in the non surgical management, as the fabrication of the vaginal dilator is done by the prosthodontist. Surgery is another mode of treatment following which prosthetic dilators are still necessary to prevent vaginal stenosis.

CASE REPORT

A 25 year old female was referred to the Department of Prosthodontics, at Faculty of Dental Sciences, Porur, Chennai, India, from the Department of Obstetrics and Gynaecology, Sri Ramachandra University, Porur, Chennai, India for the fabrication of a prosthetic vaginal dilator. The clinical examination was carried out by the gynaecologist along with the prosthodontist. The patient was diagnosed with complete Müllerian agenesis karyotype 46xx, with normal secondary sexual characteristics. A clinical examination revealed a vaginal opening measuring 0.75 cm in diameter. The diagnosis, treatment plan, duration of the treatment as well as the cost were explained and discussed with the patient and her attendee. The treatment plan was to gradually dilate the vaginal opening to allow normal sexual function. To accomplish this, customised acrylic resin stents were made, with increasing measurements over a period of four months. After procuring a written consent from the patient, the treatment was commenced.

FABRICATION

The measurements obtained were sent to the Department of Prosthodontics. A wax pattern (Hindustan Modelling Wax, India) was fabricated which was flasked, dewaxed, and processed in acrylic resin (DPI, Heat cure). The prosthesis was cylindrical, narrowing on the top to allow ease in insertion and a base to help in the removal. The fabricated prosthesis was trimmed, finished and highly polished to achieve an even and smooth surface.

During the first session the patient was given a vaginal dilator measuring 2.5 cm in length and 1cm in width. The patient was advised to use the prosthesis with the help of a lubricant Kyjelly to allow easy and less painful insertion for duration of 4-5 hours a day for a period of one month. Retention and pressure was applied and maintained by the use of tight underwear. She was also instructed to maintain good personal hygiene and of that of the stent. A thorough wash under fast running water and cleansing with a mild detergent was advised before and after usage to prevent fungal growth. On reviewing after a period of one month the patient was found to be using the dilator with comfort. Sequentially the measurements were increased to (length x width) 3.5 x 1.5cm, 4 x 1.5cm and finally 7.5 x 2cm over a period of three months. The initial prosthesis was relined to achieve the new measurement. To achieve this, the prosthesis was roughened by the use of sand paper, then covered by wax till the desired measurements of 3.5x1.5cm were achieved, then it was flasked, dewaxed and processed (Fig 1). The final relined prosthesis was then trimmed, finished and highly polished (Fig 2). As per the treatment plan, the patient was reviewed after a period of one month and given a newly relined prosthesis measuring 4x1.5cm (Fig 3). After a total of four months the patient was given a final prosthesis measuring 7.5x2cm (Fig 4). This was used for another month. On reviewing, the patient was found to have a vaginal opening measuring 7.5x2cms.

Fig 1. Wax pattern fabricated measuring 2.5 x 1.5 cm



Figure 2Fig 2. Finished prosthesis



Figure 3 Fig 3. Relined prosthesis



Figure 4Fig 4. Final prosthesis



DISCUSSION

Müllerian agenesis is a rare condition being the second most common cause for primary amenorrhea^[2]. Müllerian agenesis is the cause for 15% of the cases of primary amenorrhea^[15]. Amenorrhea, which is the absence of menses, may be temporary or permanent in some cases. It is classified as primary or secondary amenorrhea. Primary amenorrhea is the absence of menarche up to the age of 16 years and secondary amenorrhea refers to the absence of more than 3 cycles of menses.^{[6] [16]}.

There are several treatment options to treat Müllerian agenesis. Primarily, it is important that the patient and

family undergo counselling so as to educate the patient and motivate them for treatment. The treatment options include the treatment of utero vaginal aplasia. This may either be a surgical or non surgical method, but the chosen method needs to be tailored to the individual's needs^[17]. The first procedure consists of the creation of a new cavity by a surgical or non surgical method. The second method is the vaginal replacement with a pre existing canal lined with a mucous membrane such as a segment of the bowel. The non surgical creation of a neovagina involves the non surgical dilator expansion methods that have been in use for decades. Frank's dilator method [1938] is the most commonly used non surgical procedure^[15]. Frank's hand held technique had technical limitations such as fatigue and uncomfortable positioning^{[18] [19]}. Ingram's [1981] variation of this method was the bicycle stool method which provided perineal pressure via the narrow and elevated anterior portion of the bicycle seat^[18]. Robert C. P conducted a study using Ingram's technique of dilation. The difficulties faced in this method were the fabrication of the bicycle in a busy outpatient clinic [20]. In the year 2006 Mee-Hwa Lee modified Ingram's technique by using an ordinary chair^[21]. The technique used in the management of these patients was a modification of the Frank's handheld method. The material used for the fabrication of the dilator was acrylic resin which allowed relining as per the requirement when compared to Pyrex that was used by Frank. The advantage of the technique mentioned in this article was the use of tight underwear which applied firm, continuous and constant pressure to the vaginal tissue without the use of the patient's hands.

There are various surgical methods for the creation of a neovagina such as the Abbe- McIndoe operation, The Vecchietti operation and the sigmoidal coloplasty. The Abbe- McIndoe operation involves the dissection of a space between the rectum and the urinary bladder and the placement of a mould covered with skin graft, followed by the use of a prosthetic vaginal dilator. The Vecchietti operation is a mixture of surgical and non surgical procedures. It involves the creation of a neovagina via dilation with a traction device attached to the abdomen, and following sutures and the placement of a plastic olive in the vaginal dimple^[1]. The Sigmoidal coloplasty involves the replacement or creation of a vagina by grafting a 12-18 cm long segment of sigmoid^[22]. It is believed to be an efficient procedure giving excellent results, although complete adequacy for coital function often requires prolonged care and support[23].

The non surgical creation of a neovagina with the help of prosthetic vaginal dilators is usually the first line of treatment if suitable^[1]. Dilators are often helpful in widening a narrowed vagina, without resorting to surgery. It is a preferred mode of treatment as it is a more affordable method. Compared to surgical methods, non surgical vaginal dilation has the advantage of low morbidity, the creation of a more physiological milieu and no surgical scarring^[21]. The vaginal tissues become softer and more pliable over a period of time, gradually developing in regions previously undeveloped or where it is rather narrow. Sitting on a race bicycle seat stool is an alternative for vaginal dilatation [6] [16]. Surgical treatment should be considered only when the patient can participate in the decision making wishes to become sexually active and is highly motivated to use a vaginal prosthesis for several months after surgery [24][25]. Following vaginal surgery, it may also be necessary to use dilators to keep the tissues around the operation site healthy.

Over the years various studies have shown success in patients with MRKH syndrome being treated by the non surgical approach. Costa et al (1997) [21] [26] reported 87- 91%, Robson and Oliver (2000) [21] 27] reported 64%, and Robert C P (2001) [20] [21], more than 90% were able to achieve anatomic and functional success by vaginal dilatation [6] [20].

Prosthetic vaginal dilators made with acrylic resin tend to be heavier than those made with silicon material. Acrylic resin is a harder material as compared to silicon. Silicon though a softer material, due to its flexible property over acrylic resin, may cause a failure to achieve the desired size of the vagina. Also considering the patients financial background, acrylic resin was the material of choice. The fabrication of an acrylic resin stent is less tedious, as it also permits relining, hence reducing expense. It is also easy to keep clean, preventing any deposition, allowing for better hygiene.

CONCLUSION

The vaginal tissue is an elastic tissue that has the tendency to expand during the insertion of a dilator. However, it takes several months to achieve this goal. The 25 year old female patient who was referred to the Department of Prosthodontics, Faculty of Dental Sciences, Porur, Chennai, India was reviewed after a period of four months. She was found to have a vaginal opening measuring 7.5cm in length and 2 cm in width. The desired measurements were achieved economically with the help of a highly polished acrylic resin dilator. The patient reported back for a routine examination one month after the completion of her treatment. External

examination revealed a satisfactory vaginal opening, and her personal history revealed a comfortable sexual life with her husband.

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