

# Megameatus Intact Prepuce: Families May Find it Difficult to Give A Consent to Surgery in Non-Severe Cases, and the Surgery Can be More Difficult in Late Presentation

## Megameatus İntakt Prepisyum: Aileler Komplike Olmayan Olgularda Cerrahiye Onay Vermekte Zorlanabilirken Gecikmiş Vakalarda Cerrahi Zor Olabilir

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**ABSTRACT Objective:** We presented rare 8 megameatus intact prepuce (MIP) hypospadias variants and perspectives of families and patients regarding the surgery and our outcomes. **Material and Methods:** The 8 glanular MIP cases were admitted to our clinic between 2020-2023, aged between 3 to 17. The 6 cases were presented for circumcision, 2 were admitted with self suspicions. A 17-year-old was admitted for abnormal meatal appearance, considering self-awareness in advanced age; an 11-year-old family had suspected an anomaly after circumcision. Meatuses were glanular in 5, coronal in two, and distal penile in one. Mathieu flap techniques for uncircumcised and meatal advancement and glanuloplasty (MAGPI) for circumcised cases were preferred. **Results:** In four cases, the family declined to have the operation, due to a perceived lack of abnormalities. Two uncircumcised MIP cases were successfully treated using Mathieu flap techniques. The one circumcised glanular MIP was repaired by the MAGPI technique, resulting in late fistula formation. The 17-year-old case was treated with Mathieu techniques using prepuce flaps, but ended up with a late fistula formation due to unpreventable erection episodes. **Conclusion:** MIP is a rare hypospadias variant and is commonly concealed by the foreskin causing difficulty in detection, delayed diagnosis, sometimes even after circumcision, and may cause families to reject surgical correction due to a perceived lack of abnormalities. However, as it is shown in our series, a lack of preputial tissue or advanced age may negatively impact the operation's success. Families should be informed about the surgical difficulty in late presentation. The Mathieu techniques can be safely applied in non-circumcised MIP cases.

**Keywords:** Megameatus intact prepuce; family perspective; hypospadias; Mathieu technique

**ÖZET Amaç:** Biz bu çalışmada, nadir görülen 8 megameatus intakt prepisyum (MİP) hipospadias varyant vakasını, ailelerin ve hastaların cerrahiye bakış açısını ve sonuçlarımızı sunmak istedik. **Gereç ve Yöntemler:** Sekiz MİP vakası kliniğimize 2020-2023 yılları arasında başvurdu, yaşları 3-17 arasında değişmekte idi. Altı vaka sünnet talebi ile başvurdu, 2 vaka kendi şüpheleri üzerine başvurdu; 17 yaşında olan abnormal meatus görünüşü ile ki ileri yaşta kendi farkındalığını düşündürüyor, 11 yaşındaki vakanın ise ailesi sünnet sonrası anormallikten şüphelenmişti. Üretral açılım 5 vakada glanular, 2 vakada koronal ve bir vakada distal penildi. Sünnetsiz vakalarda Mathieu flap tekniği, sünnetli vakalarda ise meatal advancement and glanuloplasty (MAGPI) tekniği kullanıldı **Bulgular:** Dört vakada aile cerrahi önerisini kabul etmedi, sebep olarak görünür ve fonksiyonel bir anomali olduğunu düşünmediğini belirtti. İki sünnet olmamış vaka, Mathieu flap yöntemi ile başarılı bir şekilde tedavi edildi. MAGPI yöntemi ile onarılan bir glanular MIP vakasında geç fistül gelişti. 17 yaşındaki Mathieu ve prepisyum flap yöntemi ile onarılan olgu, önlenemez ereksiyonlar nedeni ile geç dönem fistül oluştu. **Sonuç:** MİP nadiren görülen bir hipospadias varyantı olup, genellikle sünnet derisi ile gizlenmiştir. Bu durum tanıda gecikmeye, sünnet sonrası tam konulmasına veya görünümün ve işlevin normal olduğu düşüncesi ile ailelerin cerrahi önerisini kabul etmemesine neden olabilir. Bununla birlikte serimizde de gösterildiği gibi sünnet derisinin olmayışı ve ilerlemiş yaş cerrahi başarısını negatif etkileyen faktörler olabilmektedir. Aileler geciken başvurulardaki düşük cerrahi başarısı hakkında mutlaka bilgilendirilmelidir. Sünnetli olmayan vakalarda Mathieu tekniği güvenli bir şekilde uygulanabilmektedir.

**Anahtar Kelimeler:** Megameatus intakt prepisyum; aile bakış açısı; hipospadias; Mathieu tekniği

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Observing the glans at the ventral faces of the penis in uncircumcised boys suggests hypospadias. Quite the contrary, sometimes doctors and, more often, family overlook hypospadias in uncircumcised boys with intact prepuce. The prepuce may be excised during circumcision incorrectly, causing difficulty in the later operation. The scenario can be more complex in partial megameatus intact prepuce (MIP) cases due to the lack of apparent functional abnormality, and the family smoothly denies the surgery. This study aims to demonstrate seven MIP cases and the families' perceptions.

## MATERIAL AND METHODS

Following the obtaining of Ümraniye Training and Research Hospital Ethics Committee approval (date: August 10, 2023, no: B.10.1.TKH.4.34.H.GP.0.01-265) patients were retrospectively evaluated. It is stated that all studies including the "human" element were conducted in accordance with the 2008 principles of the Declaration of Helsinki (<https://www.wma.net/policies-post/wma-declaration-of-helsinki-ethical-principles-for-medical-research-involving-human-subjects/>). The 8 mega MIP cases were presented to our clinic between 2020-2023 (Table 1). The cases was incomplete form in 7 and complete form in 1. The age range was between 3-17 years old. In 7 cases, the preputium covered the glans completely; in one case, there was a ventral preputial developmental defect (Figure 1). 6 cases were admitted for circumcision, and MIP formation was diagnosed after the examination. Two cases presented with suspicion of penile meatal abnormality, a

17-year-old boy presented by a self-suspicion, and the examination revealed a complete MIP; in this case, the sulcus was very large, causing a bilobed glandular appearance, the other, a ten-year-old boy had been circumcised two years ago, and the suspicion of having anomalies raised after circumcision and cause a doctor visit (Figure 2a, Figure 2b, Figure 3). These two cases were an example of self-awareness of megalomeatus abnormality. The urethral opening was located in the coronary sulcus in 3 cases, in the distal penile in 1 case, and in the glanular in 4 cases (Figure 1, Figure 4).

Families of 4 cases did not accept MIP operation after receiving the diagnosis, stating that there is no appearance penile shape abnormality or any urination problem in their children. They demanded circumcision surgery only. The family was fully informed about the anomaly and possible late presentation scenarios.

## RESULTS

Mathieu flap techniques was preferred in three MIP uncircumcised cases. A meatal-based penile flap was folded toward the glance and sutured to the urethral plate after creating wound edges on it. Ombredanne and Byars's flaps covered the created ventral penile defects (Figure 5). The meatal advancement and glanuloplasty (MAGPI) technique was the preferred technique for the circumcised ten years old boy. 6/0 absorbable thread was used for suturing, and urinary catheters were taken on seven days postoperatively.

We found the Mathieu technique combined with preputial flaps is helpful in two non-circumcised

TABLE 1: Characteristics of presented megameatus intact prepuce cases.

Age	Meatus location	Preputium	Operation	Family consent	Self-awareness	Complication
3	Incomplete glanular	Intact	-	No	No	-
5	Incomplete glanular	Intact	-	No	No	-
6	Incomplete coronal	Intact	-	No	No	-
6	Incomplete coronal	Intact	-	No	No	-
6	Incomplete distal	Intact	Mathieu	Yes	No	No
10	Incomplete glanular	Circumcised	MAGPI	Yes	Yes	Fistula
13	Incomplete coronal	intact	Mathieu	Yes	No	No
17	Complete glanular	Intact	Mathieu	Yes	Yes	Dehiscence

MAGPI: Meatal advancement and glanuloplasty.



**FIGURE 1:** The prepuce is seen as partially circumcised due to incomplete development of the prepuce on the ventral face of the glans in a 6 year old megameatus intact prepuce case.



**FIGURE 3:** Circumcised megameatus intact prepuce appearance in a 10 year old boy.



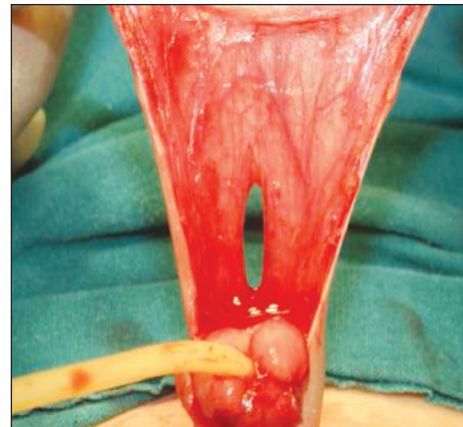
**FIGURE 2a:** A picture of a completely developed prepuce in a 17-year-old case



**FIGURE 4:** A coronal opening meatus in a 13 year old boy.



**FIGURE 2b:** A picture of a complete mega meatus a bilobed glandular appearance.



**FIGURE 5:** Ombredanne flap is a dorsal prepuce flap that contains a hole in the middle of the island, likened to its first letter.

cases. The patients experienced satisfactory post-operative results and no complications during a year-long follow-up visit. Their urinary flow was normal.

A 17-year-old boy was operated on with the same technique, but a suture line dehiscence occurred on the fifth day after surgery, caused by uncontrollable erection episodes. Although it closed adequately, a fistula formed during the two-week follow-up visit, and the patient refused a further fistula repair surgery.

In a 10-year-old circumcised boy, for which we preferred the MAGPI technique, a fistula formation was noticed on the suture line after urinary catheter removal on the fifth day. A waterproofing interposition layers technique was used for fistula repair, and no complication occurred during the last six months' follow-up visit.

## DISCUSSION

MIP is a rare form of hypospadias in 1-3% of cases.<sup>1</sup> However, the actual number of cases may be higher than reported because some people may have the condition without realizing it or decline treatment after diagnosis.<sup>2</sup>

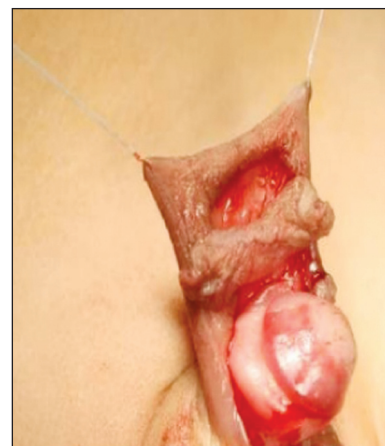
A typical appearance is like an entirely developed preputium, wide fish mouth opening urethra, shovel-shaped glans, no ventral curvature, and a wide fossa navicularis (Figure 2a, Figure 2b). The condition was first described by Juskiewenski et al. as glandular placed hypospadias, later evaluated in detail by Duckett and Keating, and the Pyramid procedure was offered as an effective surgery for it.<sup>3,4</sup> According to the urethral opening, it is further classified into three groups: glandular, coronal, and distal.<sup>5</sup> New publications have made reaching more extensive series and surgical options possible.

Some families may choose not to bring their children for visits due to uncomplicated hypospadias that is not related to curvature or does not cause urinary difficulty, even if the preputium does not completely cover the glans. The latter is known as prophets' circumcision in Islamic areas and may be perceived as usual.<sup>6</sup> When the preputium does not completely cover the glans with ventral face defect, it can easily be diagnosed earlier; however, in a fully

developed prepuce (Figure 1, Figure 2). The doctors can even miss the diagnosis in the case of completely covering preputium. It may end up with a post-circumcisional late diagnosis, as we presented in our series: 6 patients were unaware of having MIP anomaly before the examination.

In date, it is recommended that surgeons practicing on circumcision should examine the urethral meatus before the operation.<sup>7</sup> Regarding MIP cases, missed diagnosis can result in accusations after circumcision by the family. In the study Peretz and Westreich 7 MIP diagnoses out of the 21 were missed by the urologists or plastic surgeons, resulting in a file suit by the family due to an incorrect circumcision procedure.<sup>8</sup> However, with growing evidence, the importance of preputium tissue for a successful MIP operation has become a topic of debate. According to the Snodgrass and Khavari and Pieretti et al. series, circumcision before the MIP operation is believed not to affect its success.<sup>9,10</sup> Nonetheless, we believe that the surgeon's level of experience can contribute these results, and the presence of prepuce tissue has an impact for successful operation.

The preputium can be used for supporting the urethral plate in hypospadias surgery (Figure 5, Figure 6). For this purpose, different preputial flaps have been described. In Ombredane flap, a hole is opened at the visceral preputium's center and put throughout the glans to cover the neourethra like an apron (Figure 5). In Byras flap, the preputium is sep-



**FIGURE 6:** Preputium tissue can be used for either a skin graft or flap for hypospadias surgery.

arated into two island flaps and brought to the neourethra bilaterally. We used these two different preputial flaps with great success, indicating that having prepuce tissue may lead to a good achievement in the MIP operation.

We believe that the fact that four families have refused surgery, which comprises half of our patient group, is subject to considerable debate. Family's reluctance to MIP surgery is understandable, considering an ongoing debate in the literature about which MIP cases require surgery.<sup>11</sup> It is still uncertain whether surgical intervention is necessary for patients with partial MIP who have normal urinary and sexual function, and whose daily life is not affected by the condition.<sup>12</sup> However, in light of increased awareness regarding cosmetic appearance, a surgical correction can be offered to the family to prevent late readmission which may result in difficulty in surgery.<sup>2</sup> On the other hand, surgery can safely be offered in patients with a meatal opening close to or below the coronal margin, a deep glans cleft, a broad, splayed-out glans, and urinary difficulty caused by the anomaly. Only one case had self-awareness of having a urethral meatus abnormality. Our two late presented cases were a typical example for encountered difficulty for successful operation either by being a late presentation or lack of preputial tissue, so we strongly suggest that families hesitating about surgery must be fully informed about the issue and consequences of possible late presentation.

The goal of MIP treatment is to decrease the size of the large urethral opening and relocate it to the center of the glans. A number of technics have been offered for MIP in the literature and increasing number of articles have focused on this rare hypospadias variant.<sup>5</sup> Some surgeons have offered MAGPI, or Glanular approximation operation, for glanular cases. On the other hand, for coronal, and distal penile type MIP the Mathieu, Duplay, or tubularized incised plate technics can be offered.<sup>13</sup> In Mathieu technics, the reverse proximal urethral flap is used to support the neo-urethra to avoid urethral stricture formation (Figure 5). The tubularized urethral plate urethroplasty (TIPU) operation has also been offered for the distal cases.<sup>12</sup> In a 27 cases series study, TIPU was preferred for 13, Duplay for 7, Meathieus for 5, and

MAGPI for 1 case, and no differences between the techniques were shown.<sup>14</sup> In a 13-case retrospective series, 12 cases were operated with the TIPU technique. For one glanular case, the MAGPI was preferred, and no complication was encountered during the follow-up.<sup>9</sup> We preferred the Mathieu technique in our intact preputial cases due to take extra advantages of preputium; a year follow-up was event-free and the MAGPI technique for a glanular circumcised case was successful.

## CONCLUSION

Even if our presented study consists of limited cases series, it is valuable in reflecting the family's perspective on surgery and reasons for this. It also clearly showed that the success of Mathieu techniques in this type of hypospadias variant is paramount, and the consequences of late presented MIP cases are fairly appreciable. However, to gain a wide comprehensiveness to make a more precise recommendation to the families about their decision and the long-term consequences of this particular hypospadias variant, more case series studies are needed.

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### Conflict of Interest

*No conflicts of interest between the authors and / or family members of the scientific and medical committee members or members of the potential conflicts of interest, counseling, expertise, working conditions, share holding and similar situations in any firm.*

### Authorship Contributions

**Idea/Concept:** Timuçin Şipal, Ferhat Yakup Suçeken; **Design:** Timuçin Şipal, Ferhat Yakup Suçeken; **Control/Supervision:** Timuçin Şipal, Ferhat Yakup Suçeken; **Data Collection and/or Processing:** Timuçin Şipal, Ferhat Yakup Suçeken; **Analysis and/or Interpretation:** Timuçin Şipal; **Literature Review:** Timuçin Şipal; **Writing the Article:** Timuçin Şipal; **Critical Review:** Timuçin Şipal; **References and Fundings:** Timuçin Şipal, Ferhat Yakup Suçeken; **Materials:** Timuçin Şipal, Ferhat Yakup Suçeken.

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