The association of meatal stenosis and infant circumcision

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ABSTRACT

Background. The association of meatal stenosis with age at circumcision is controversial. We noticed a high rate of meatal stenosis in a region where early circumcision is traditional. The aim of this study is to compare the age at circumcision between boys with or without meatal stenosis.

Methods. After ethical approval, families of children with meatal stenosis were questioned about age at circumcision and reason for circumcision. Control group consisted of patients with diagnoses other than penile abnormalities, a normal urethral meatus, and having no symptoms about urination. Patients with a history of therapeutic circumcision were excluded from the study.

Results. Between November 2016 and November 2020, 115 patients with meatal stenosis were admitted. All were corrected with ventral meatotomy under general anesthesia. Median age at circumcision was 3 (min:0-max:111) monthsand age at admission was 74 (min:22-max:194) months. Control group consisted of 205 boys. Median age at circumcision was 5 (min:0-max:122) months and age at admission was 96 (13-202) months. There was a statistically significant difference between groups in terms of age at circumcision (p=0.024) but none for age at admission (p=0.356). There was a twofold increase in the meatal stenosis rate (39% vs. 23%) if circumcision was performed before age one (p=0.018). There was no difference between the patients circumcised in the newborn period and later (38% vs. 36%, p=0.778).

Conclusions. Our study supports the previous reports suggesting a relation of risk for meatal stenosis and age at circumcision and presents data that age one might be a cutoff for this risk.

Key words: circumcision, infant, neonatal, meatal stenosis, meatal web.

American Academy of Pediatrics (AAP) changed its policy statement on circumcision in 2012, and specified that the benefits of circumcision in the newborn period are sufficient to justify leaving the decision to the parents but not great enough to recommend routine circumcision for all male newborns. Since then, infant circumcision has preserved its popularity.

Meatal stenosis is one of the common complications of circumcision.² Meatal stenosis

rate is higher in circumcised males^{3,4} but the association of meatal stenosis with age at circumcision is controversial. We noticed an extraordinarily high rate of meatal stenosis in a region where early circumcision is traditional. The aim of this study is to compare the age at circumcision between boys with or without meatal stenosis.

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Material and Methods

The study was mainly based on retrospective inquiry of the parents, but also included physical examination and voiding observation of the children. The inquiry consisted of two short questions as age at circumcision and the reason for circumcision.

The study group included patients operated due to urethral meatal stenosis between November 2016 and November 2020. The diagnosis of meatal stenosis was made observing the prolonged and upward directed urinary stream with narrow caliber. All patients with meatal stenosis were treated with meatotomy. Meatotomy was performed under general anesthesia and involved incision and suturing of the web on the ventral aspect of the meatus while calibrating the urethra using a stent with a diameter appropriate for age. None of the authors prefer dilations for meatal stenosis as they think it does not provide a permanent solution.

The control group was constituted from circumcised boys who were admitted to the same hospital between November 2019 and November 2020. These were patients with diagnoses other than penile abnormalities, having a normal-looking urethral meatus, and reporting no symptoms about urination. After informed consent, their parents were inquired about age at and reason for circumcision. Normal urine flow was confirmed with videos of voiding in the control group. Patients with a history of therapeutic circumcision and the ones with a duration less than one year since circumcision were excluded from the study.

This study was conducted at a 665 bed, secondary care children's referral hospital with approximately 485.000 patient admissions annually. The city where the study was performed is in south-east of Türkiye where early circumcision (after the 40th day of life) is traditional but not consistently performed by all. Ethical approval was obtained from the local ethics committee (Gaziantep University, Ethical Board for Clinical Studies, decision number: 2019/287). Written informed consent was obtained both from the parents and children.

The statistical analyses were performed using IBM SPSS Package Version 25 (Armonk, NY, USA: IBM Corp.). Histograms and the Kolmogorov-Smirnov test were performed

to check the normality of distribution of the continuous variables. Descriptive statistics were used to summarize patient characteristics. A p-value below 0.05 was considered statistically significant. The Mann-Whitney U test was used to compare age at circumcision and age at admission between the patients with or without meatal stenosis. Pearson Chi-square test was used to evaluate the difference in meatal stenosis rate between subgroups according to age at circumcision. A power analysis using G*power 3 software was also performed.⁶

Results

In total, 115 patients with meatal stenosis were admitted to our hospital during the study period. All were corrected with ventral meatotomy under general anesthesia, and all accepted to be included in the study. There were 205 participants in the control group. Main characteristics of the study group are present in Table I. There was a statistically significant difference between groups in terms of age at circumcision (p=0.024) but none for age at admission (p=0.356). There was also no difference between groups regarding duration between the circumcision and admission (p=0.141).

We then evaluated how meatal stenosis ratio changed in our study group regarding circumcision at different specific time periods such as during infancy, during newborn period, before the end of mini puberty, or during mini puberty (Table II). There was no difference between the patients circumcised in the newborn period and later (38% vs 36%, p=0.778). Meatal stenosis rate seemed to be higher in the first six months (Fig. 1), but the most prominent cut-off was at age 1 years. The number of patients with meatal stenosis was almost double when patients who had circumcision before age one or later were compared (39% vs. 23%) (p=0.018). There was also a statistically significant difference between the patients circumcised before or after 6 months (42% vs 27%, p=0.008) but none

Table I. Main characteristics of the study group.

	Number of patients	Age at circumcision	Age at admission	Duration between circumcision and admission
Patient Group	115	3 (0-111)*	75 (22-194)*	71 (7-182)*
Control Group	205	5 (0-122)*	96 (13-202)*	81 (12-177)*
		p=0.024	p=0.356	p=0.141

^{*}depicted as months; median (range).

Table II. Number of subjects with meatal stenosis in different time limits.

Number (9/) of subjects	Number (%) of subjects			
Number (%) of subjects —	with meatal stenosis without meatal stenosis		р	
when newborn	26 (38%)	(0.		
later	89 (36%)			
first 6 months	nonths 83 (42%) 117 (58%) later 32 (27%) 88 (73%)		0.008	
later				
first 12 months	101 (39%)	157 (61%)	0.018	
later	14 (23%)	48 (77%)		
between 0-6 months	83 (42%)	117 (58%)	0.171	
between 6-12 months	18 (31%)	40 (69%)	0.171	
during mini-puberty (2-6 months)	57 (44%)	74 (56%)	0.024	
any other time	58 (31%) 131 (69%)		0.024	

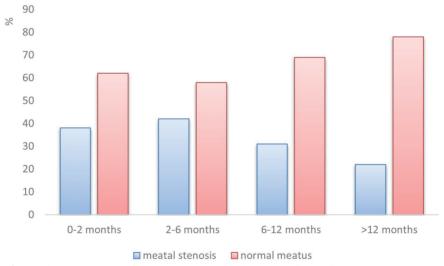


Fig. 1. The percentage of patients with meatal stenosis for each age group.

between patients circumcised in the first six months and between 6 to 12 months (42% vs 31%, p=0.171). We also evaluated circumcisions during mini puberty (2-6 months) or another time (including the newborn period), and saw a

similar finding (44% vs 31%, p=0.024). Overall, the ratio of patients with meatal stenosis seemed to be similar until age one years, and significantly smaller after then.

Discussion

Circumcision is one of the oldest and most commonly performed procedures controversy on its necessity and when to perform it continues.7 Our study focuses on the association of meatal stenosis and age at circumcision. The advantage of circumcision regarding decreased risk of urinary infections is more prominent when performed in the newborn period.8 On the other hand, the prepuce cannot be retracted fully in 96% of infants9 which necessitates forceful retraction of the prepuce during circumcision in the newborn period. Exposure of delicate mucosa to ammonium or mechanical trauma in a child with diapers, ischemia of the meatal mucosa stemming from damage to the frenular artery, and forceful degloving of the preputium have all been implicated in the etiology of meatal stenosis. 5,10,11 Therefore, an association between meatal stenosis and age at circumcision seems straightforward.

One problem while discussing meatal stenosis is its definition. The rate of meatal stenosis is highly varying among papers.^{3,11} A metanalysis reported data supporting increased risk of meatal stenosis following circumcision (with an odds ratio of 3.20) but that overall rate is low (<1%).³ On the other hand, a large retrospective series on infant circumcision revealed that one fourth of the revision surgeries were due to meatal stenosis after neonatal circumcision.¹² This is probably due to the differences in its definition.

Özen et al.¹³ reported that patients admitted as meatal stenosis following infant circumcision had a web-like structure on the ventral aspect of the meatus. Therefore, they suggested using the name "meatal web" instead of "meatal stenosis". We agree with their observation regarding the anatomy of the pathology but preferred to use the common nomenclature as their proposal did not find widespread usage. Lichen sclerosus is also associated with meatal stenosis¹⁴ which is probably a different entity. And as discussed, the effect of inflammation on

meatal stenosis is also debated. Therefore, we excluded all cases who underwent a therapeutic circumcision to understand the relationship with age solely.

Several large-scale studies showed circumcision increases the risk of meatal stenosis3, but scarce studies addressed its relation with age at circumcision and the threshold is varying. A prospective cohort study on 1100 participants by Howe¹⁵ showed that all children with meatal stenosis were circumcised neonatally. Acimi et al.16 showed a twofold increase when comparing first week and that between 7-12 months. Likewise, Machmouchi et al.11 found a higher rate of meatal deformity (reminding our definition of meatal stenosis) comparing the neonatal period with 5 months (90% vs. 11%). We found that age at circumcision was significantly smaller in children with meatal stenosis but unlike previous studies, there was no difference in the ratio of patients with meatal stenosis between the newborn period and later. It changed significantly at age one. We also searched for a specific relationship about circumcision during or before the completion of mini puberty, but the rate was similar until age 1 years with no particular association with mini-puberty. Our results may support the retractability theory for the etiology of meatal stenosis following circumcision as retractability rate of the prepuce increases to 50% at age one years9 but more data is obviously required to draw clearer conclusions.

The major limitation of our study is not demonstrating an overall meatal stenosis prevalence in the population it was performed. But our study does not make any claims regarding the prevalence or etiology of meatal stenosis. Another problem is that the majority of the patients were circumcised in infancy, which we think can also explain the high number of patients with meatal stenosis in a study involving patients in a four-year period. Also, we do not know when meatal stenosis exactly happens, therefore we do not know if the participants in the control group will experience meatal stenosis later. Participants who were admitted

in the year following circumcision were excluded from the study to overcome this bias. Another limitation is that our study involves no data about the technique of circumcision, postoperative care after the circumcision or preservation of frenulum which can also be contributive factors for meatal stenosis. Besides these limitations of a retrospective study, the data retrieved is retrospective but what we ask the parents is their child's age at circumcision and if it was due to therapeutic reasons, which we think are not questions open to recall bias.

As mentioned above, meatal stenosis definition and rate differed significantly among the published papers, so a proper estimation of sample size was also impossible. Minimum sample size for each group had to be somewhere between 4 and 7178 according to published papers.3,11 Therefore, we aimed to include all patients with meatal stenosis and a higher number of participants in the control group (circumcised patients with no meatal stenosis). Then, we performed a post-hoc power analysis. The power of our study to detect the difference between meatal stenosis rate in patients who underwent circumcision before or after age one with a 5% level of significance was calculated as 97%.

American Association of Pediatrics leaves the decision of infant circumcision to the parents and the responsibility of informing them about its advantages and disadvantages to their physicians. The discussion with parents is mainly based on the risk of UTI or the rare catastrophic complications of circumcision. The risk for meatal stenosis is seldom discussed in detail with parents. We think it's important for a parent to know that circumcision can result in one more intervention and this actually might be associated with the age it was performed.

Our study supports previous reports suggesting a relation of risk for meatal stenosis and age at circumcision and presents data that age one years might be a cutoff for this risk. Further studies are required to investigate this association, and families should be informed about the risk of meatal stenosis while discussing timing of circumcision.

Ethical approval

Ethical approval was obtained from the local ethics committee (Gaziantep University, Ethical Board for Clinical Studies, decision number: 2019/287). Written informed consent was obtained both from the parents and children.

Author contribution

The authors confirm contribution to the paper as follows: study conception and design: ST; data collection: ST, YI; analysis and interpretation of results: ST, YI; draft manuscript preparation: ST, YI. All authors reviewed the results and approved the final version of the manuscript.

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Conflict of interest

The authors declare that there is no conflict of interest.

REFERENCES

- 1. American Academy of Pediatrics Task Force on Circumcision. Circumcision policy statement. Pediatrics 2012; 130: 585-586. https://doi.org/10.1542/ peds.2012-1989
- Weiss HA, Larke N, Halperin D, Schenker I. Complications of circumcision in male neonates, infants and children: a systematic review. BMC Urol 2010; 10: 2. https://doi.org/10.1186/1471-2490-10-2
- 3. Morris BJ, Krieger JN. Does circumcision increase meatal stenosis risk?-A systematic review and meta-analysis. Urology 2017; 110: 16-26. https://doi.org/10.1016/j.urology.2017.07.027
- 4. Berry CD, Cross RR. Urethral meatal caliber in circumcised and uncircumcised males. AMA J Dis Child 1956; 92: 152-156. https://doi.org/10.1001/archpedi.1956.02060030146007

- Langer JC, Coplen DE. Circumcision and pediatric disorders of the penis. Pediatr Clin North Am 1998; 45: 801-812. https://doi.org/10.1016/s0031-3955(05)70046-8
- Faul F, Erdfelder E, Lang AG, Buchner A. G*Power 3: a flexible statistical power analysis program for the social, behavioral, and biomedical sciences. Behav Res Methods 2007; 39: 175-191. https://doi. org/10.3758/bf03193146
- Robson WL, Leung AK. The circumcision question. Postgrad Med 1992; 91: 237-244. https://doi.org/10.1 080/00325481.1992.11701325
- 8. Morris BJ, Wiswell TE. Circumcision and lifetime risk of urinary tract infection: a systematic review and meta-analysis. J Urol 2013; 189: 2118-2124. https://doi.org/10.1016/j.juro.2012.11.114
- 9. Dewan PA, Tieu HC, Chieng BS. Phimosis: is circumcision necessary? J Paediatr Child Health 1996; 32: 285-289. https://doi.org/10.1111/j.1440-1754.1996. tb02554.x
- Morris BJ, Moreton S, Krieger JN. Meatal stenosis: getting the diagnosis right. Res Rep Urol 2018; 10: 237-239. https://doi.org/10.2147/RRU.S192310

- 11. Machmouchi M, Alkhotani A. Is neonatal circumcision judicious? Eur J Pediatr Surg 2007; 17: 266-269. https://doi.org/10.1055/s-2007-965417
- 12. Pieretti RV, Goldstein AM, Pieretti-Vanmarcke R. Late complications of newborn circumcision: a common and avoidable problem. Pediatr Surg Int 2010; 26: 515-518. https://doi.org/10.1007/s00383-010-2566-9
- 13. Özen MA, Gündoğdu G, Taşdemir M, Eroğlu E. Complication of newborn circumcision: meatal stenosis or meatal web? J Pediatr Urol 2017; 13: 617.e1-617.e4. https://doi.org/10.1016/j.jpurol.2017.05.007
- Homer L, Buchanan KJ, Nasr B, Losty PD, Corbett HJ. Meatal stenosis in boys following circumcision for lichen sclerosus (balanitis xerotica obliterans). J Urol 2014; 192: 1784-1788. https://doi.org/10.1016/j. juro.2014.06.077
- 15. Van Howe RS. Incidence of meatal stenosis following neonatal circumcision in a primary care setting. Clin Pediatr (Phila) 2006; 45: 49-54. https://doi.org/10.1177/000992280604500108
- Acimi S, Abderrahmane N, Debbous L, et al. Prevalence and causes of meatal stenosis in circumcised boys. J Pediatr Urol 2022; 18: 89.e1-89. e6. https://doi.org/10.1016/j.jpurol.2021.10.008