ORIGINAL ARTICLE

Prevalence of acquired undescended testis in 6-year, 9-year and 13-year-old Dutch schoolboys

W W M Hack, K Sijstermans, J van Dijk, L M van der Voort-Doedens, M E de Kok, M J Hobbelt-Stoker

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See end of article for
authors' affiliations

Correspondence to:
Dr W W M Hack, Medical
Centre Alkmaar, Department
of Paediatrics,
Wilhelminalaan 12,
Alkmaar 1815 JD, The
Netherlands;
w.hack@mca.nl

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Objective: To investigate the prevalence of acquired undescended testis (UDT) in Dutch schoolboys.

Design and participants: As a part of routine school medical examinations, during a 2-year period (2001–3), testis position was determined in 6-year, 9-year and 13-year-old schoolboys. Before the examination, a parent questionnaire was sent inquiring both about the position of the testes and whether the child had been admitted earlier to hospital for orchidopexy. In 6-year and 13-year olds, a physical examination was performed by the school medical officer; in 9-year olds, a school nurse interview was held. Each boy for whom there was any doubt of the scrotal position was referred to the hospital for examination of both testes.

Setting: Institution for Youth Health Care "Noordkennemerland" and Medical Centre Alkmaar, Alkmaar, the Netherlands.

Results: Testis position was determined in 2042 boys aged 6, 1038 aged 9 and 353 aged 13. Of these, 47, 53 and 8 boys, respectively, were referred to the hospital and seen for further evaluation. The diagnosis of acquired UDT was made in 25 boys aged 6, 23 aged 9 and four aged 13. In 33 boys, a congenital UDT was diagnosed; 32 (97%) had already been diagnosed and treated at an early age.

Conclusions: The prevalence of acquired UDT for 6-year, 9-year and 13-year olds was, respectively, 1.2% (25/2042), 2.2% (23/1038) and 1.1% (4/353). In addition, congenital UDT is treated during the early years of life and, in contrast with popular belief, screening programmes for detecting UDT in the early years are successful.

At present, undescended testis (UDT) is categorised in congenital and acquired forms.^{1–3} The prevalence of congenital UDT is about 0.8–1.1% by 1 year of age,⁴ whereas orchidopexy (ORP) rates appear to be three times higher than expected if only this prevalence of congenital UDT is taken into account.^{5–6} Further, despite the recommendations for early ORP, most operations are performed late in childhood.^{7–8}

The true prevalence of acquired UDT is still unknown. There is increasing evidence from retrospective studies in selected groups of boys to suggest acquired UDT as a common occurrence, outnumbering congenital UDT by a factor of 2–3.⁹ This prospective study was initiated to provide estimates of the prevalence of acquired UDT in unselected groups of boys at various ages. The results of this study may shed light on the high ORP rate in later childhood.^{9–10}

DESIGN AND PARTICIPANTS

General design

The study was performed in the Youth Health Care Institution of the Gemeenschappelijke Gezondheidsdienst "Noordkennemerland", Alkmaar and in the Medical Centre Alkmaar. The district has 119 primary schools. During a 2-year period (2001–3), 6-year, 9-year and 13-year-old boys were studied. Routine medical examinations at school take place at these ages, and this study was part of these medical examinations. The policy of the Youth Health Care Institution is to focus more on 6-year olds. Therefore, more 6-year olds are seen annually than 9-year and 13-year olds. Six-year-old boys were born between 1 October 1995 and 1 October 1997, 9-year olds between 1 October 1992 and 1 October 1994, and 13-year olds between 1 October 1989 and 1 October 1990. In the end, 13-year olds were studied for only a 1-year period. They were initially unwilling to participate and an information campaign proved essential. Before examination, a parental information

letter informing about the study, together with a parental questionnaire, was sent inquiring about their son's testes position and whether he had previously been admitted to hospital for ORP. We did not enquire whether the operations were specifically for maldescent. Inquiries about other groin surgery as well as other associated conditions and family history were not made. At the school medical examination, the questionnaire was collected by the school nurse.

Youth Health Care Institution 6-year and 13-year olds

In boys aged 6 and 13 years, testicular examination was carried out by a school doctor as part of a routine medical programme. Scrotal examination was performed by the doctor while the boy was in a standing position, according to a previously approved protocol. A solely screening examination was performed to determine whether or not the testes were present in the scrotum.

9-year olds

Boys aged 9 years were seen by a school nurse who routinely performed audiometric assessment, tested visual acuity, and measured height and weight. Additionally, an interview was performed inquiring about, among other things, testes position, and to collect the parental questionnaire. A boy was referred if the parent reported any doubt on the scrotal position of one or both testes.

Referral

Each boy in whom there was any doubt on the scrotal position of one or both testes was referred to the hospital for further examination. Adequate referral was ensured as follows: the questionnaires were collected at regular intervals of a few

Abbreviations: ORP, orchidopexy; UDT, undescended testis

weeks at the Youth Health Care Centre by the research nurse of the hospital. At the same time, the data on previous testicular positions, determined by proficient Youth Health Care medical officers at various ages during routine medical inspections, were collected from the Youth Health Care medical records. In view of the long-term cooperation with the Youth Health Care medical officers, instructions have been repeatedly given by the authors about the definition of UDT.³ Parents were then contacted by the research nurse by telephone and requested to visit the hospital, with their son, for further evaluation. Simultaneously, an appointment for the outpatient clinic was made.

Medical Centre Alkmaar

At the outpatient clinic, a full physical examination was performed on each boy by the same paediatrician (WH). Testicular examination was carried out by a two-handed technique with the boy in the supine and cross-legged positions. All boys were seen within a few weeks of referral.

Definitions

A “retractile testis” was defined as a testis that could be manipulated into a (low) stable scrotal position where it remained until the cremasteric reflex was elicited. Traction on cord structures was not painful. An “undescended testis” was defined as a testis which could not be manipulated into a stable scrotal position and further tension on cord structures was painful.² A “high scrotal testis” was defined as a testis which can (still) be brought through the scrotal entrance into a high but unstable scrotal position, although further traction on cord structures is painful. An acquired UDT was defined as a UDT for which a previous scrotal position was documented on at least one occasion. Acquired UDT involves “high scrotal testes” and “(inguinal) ascending testes”.

Ethics approval

The study protocol was approved by the ethics committee of the hospital (reference number: MO 1-08).

RESULTS

A total of 3627 boys were invited for the school medical examination, and 3433 (94.7%) attended the routine medical inspections at school at ages 6, 9 and 13 years (table 1). In all medical examinations the questionnaire was returned, and in most cases a parent attended the school medical examination.

6-year olds

A total of 2093 boys received an invitational letter and a questionnaire and 2042 (97.6%) were seen by the school doctor. At the school physical examination, 57 non-scrotal testes were

found in 48 boys. Of those, 47 (98%) boys were seen at the hospital, and in 25 boys an acquired UDT was diagnosed. In all of these boys a previous scrotal position was documented: in two testes, at least twice; in two, at least three times; and in 24 testes, at least four times.

In two boys, a congenital UDT was diagnosed. In one, scrotal descent was documented once immediately after birth, and thereafter the testis had never been palpated. After referral, ORP was performed and an abdominal testis was found. Therefore, observer error in physical examination immediately after birth is likely, although early ascent cannot be excluded. The other boy was known to have congenital UDT from birth, but his parents refused surgical treatment.

9-year olds

At the age of 9 years, 1146 boys were invited by letter, and 1038 (90.6%) were interviewed by the school nurse. All these questionnaires were returned; 56 parents indicated that one or both testes of their son was non-scrotal; 53 (95%) boys were seen at the hospital, and in 23 boys an acquired UDT was diagnosed. In these boys, a previous scrotal position was documented at least once in two testes, at least twice in three, at least three times in three, and more than three times in the remaining 19 testes.

A congenital UDT was found in one boy. He was known to have Prader–Willi syndrome and congenital UDT from birth. On request by the parents, active treatment was not performed.

13-year olds

A total of 388 boys received an invitational letter with a questionnaire; 353 (91%) boys were seen by the school medical officer and testicular position was determined; 11 testes in eight boys were judged as non-scrotal. In four boys, the testis was diagnosed as a retractile testis. In the other four, a diagnosis of acquired UDT was made. A previous scrotal position was documented at least three times in one testis and more than three times in the remainder. In the four boys with acquired UDT, the parents were unaware of the non-descent.

The prevalence for acquired UDT in 6-year, 9-year and 13-year olds was, respectively, 1.2% (25/2042), 2.2% (23/1038) and 1.1% (4/353).

Congenital UDT

ORP had been performed for congenital UDT, by 6 years of age in 17 boys, by 9 years of age in 11 boys, and by 13 years of age in two boys. In addition, in two 6-year olds, in one 9-year old and in no 13-year olds, a congenital UDT was found. The prevalence of congenital UDT in 6-year, 9-year and 13-year olds was, respectively, 0.9% (19/2042), 1.2% (12/1038) and 0.6% (2/353). In one boy congenital UDT was misdiagnosed at birth, and in the remaining 32 (97%) the condition was well diagnosed and surgically treated ($n = 30$) or treatment advised ($n = 2$).

DISCUSSION

In this series, the prevalence of acquired UDT in 6-year, 9-year and 13-year olds was, respectively, 1.2%, 2.2% and 1.1%, whereas the rate of congenital UDT was 0.9%, 1.2% and 0.6%, respectively. This prevalence of congenital UDT is in accordance with the known prevalence of 0.8–1.1% in 1-year olds;⁴ therefore the questionnaire seems to be adequate. In addition, 32 of 33 cases of congenital UDT had already been treated surgically or diagnosed at an early age, indicating adequate medical youth surveillance.

The prevalence of UDT varies considerably depending on the design of the study (eg cohort studies are more reliable than

Table 1 Number of boys seen at school medical examination, together with the number of boys seen in hospital for undescended testis

	Number of boys (number of testes)		
	6-year olds	9-year olds	13-year olds
Seen at school medical examination	2042	1038	353
Seen in hospital	47	53	8
Retractile	20 (26)	29 (47)	4 (6)
Congenital UDT	2 (2)	1 (1)	0 (0)
Acquired UDT	25 (28)	23 (27)	4 (5)

UDT, undescended testis.
Data for individual testes included.

registry data studies) and the criteria and definitions used. Other compounding factors include racial and ethnic heterogeneity, gestational age and birth weight, seasonal variability and regional differences.¹¹ Reported UDT rates vary at 6 years from 0% to 2.6%, at 9 years from 0% to 5%, and at 13 years from 0% to 4%.^{12–15} A major factor explaining the wide variety, however, must be, at present, the recognition of congenital UDT and acquired UDT. Many studies on UDT prevalence do not distinguish between these forms.

Until now, a few dozen reports have been published on acquired UDT, and prevalence has been reported in selected groups of boys. These include boys who were referred for non-scrotal testis or in whom ORP had been performed for UDT. These studies showed that acquired UDT may outnumber congenital UDT by a factor of 2–3.

As in this study, congenital UDT is seen in 0.8–1.1% of 1-year-old boys, accounting for the early ORP peak. This study shows that the prevalence of acquired UDT ranges from 1.1% to 2.2%, and these figures correspond very closely to the late ORP rate of 2–3%. Therefore, we believe that acquired UDT may indeed account for the late ORP peak as suggested previously^{16–18} and that testicular ascent is a real phenomenon. At present acquired UDT is accepted as a new entity,¹⁹ although some claim it to be unrecognised congenital UDT.²⁰

Screening for UDT is one of the important elements of child health surveillance. However, it is generally believed that screening programmes fail to identify UDT at an early age, possibly because of the subtleties of the anatomy and difficulties inherent in examining children,²⁰ and that recommendations for early treatment are not followed.^{21–23} This series, however, shows that nearly all cases of congenital UDT are recognised early and treated as recommended, and that, after the age of 5 years, acquired forms are almost exclusively seen. In this series, only one (3%) of 33 congenital forms was misdiagnosed at birth, whereas the remaining cases were already treated (30 cases) or known (two cases). Therefore, the recognition of congenital UDT and acquired UDT explains why only about one third of all ORPs is performed in the first 2 years despite recommendations.^{7, 21, 24–28} Although ORP is routinely performed for acquired UDT in prepubertal boys, this procedure is heavily under debate. It has been shown that most acquired forms descend spontaneously in the peripubertal period with testicular volumes appropriate for age.²⁹ Therefore, ORP might be postponed until at least mid-puberty. In the studied boys, testicular position is still assessed annually. At present, in eight boys (seven 9-year olds and one 13-year old), the testis has descended spontaneously, whereas, in four cases, ORP had to be performed (one 9-year old, three 13-year olds).

It must be emphasised that this study has obvious limitations. It is not a population-based study, so we were unable to estimate the number of males with UDT in the general population. An occasional unrecognised (congenital) UDT can never be ruled out, owing to interobserver variability in the healthcare personnel. Potential errors may have been introduced by the difference in the number of boys included in the various age groups, owing to a difference in priority at the Youth Health Care Institution. In addition, 13-year olds were studied for only a 1-year period. In addition, not all boys were physically examined and the results were dependent on adequate parental participation. As previous authors have documented ascent at ages as young as 1–3 years,^{30, 31} ORP in earlier years could potentially have been performed for both congenital and acquired UDT. A population-based study is needed to fully ascertain the true incidence of acquired UDT in the general population. We speculate that this rate might be significantly higher than seen in this series.

What is already known on this topic

- At present, undescended testis (UDT) is categorised in congenital and acquired forms.
- The prevalence of congenital UDT varies from 0.8% to 1.1% at 1 year of age.
- Until now, there are no studies documenting the prevalence of acquired UDT.
- Acquired UDT may explain the high rate of orchidopexies performed later in childhood despite recommendations to perform this operation at between 6 and 24 months of age.

What this study adds

- This is the first study to report on the prevalence of acquired UDT in boys. It confirms the high rate of this phenomenon seen later in childhood and may offer an explanation for the high rate of orchidopexies performed later in childhood.
- This series shows that, in contrast with popular belief, medical youth screening programmes do not fail in detecting UDT in the early years.

CONCLUSIONS

In this series, the prevalence of acquired UDT in 6-year, 9-year and 13-year olds varies from 1.2% to 2.2%. After the age of 5 years, only acquired UDT is seen. Congenital UDT is recognised and treated in the first years of life as recommended, indicating adequate medical youth surveillance. Routine medical examinations in older boys—that is, after the age of 5 years—should also include examination of both testicles. Finally, those involved in the care of boys with UDT ought to realise that a boy referred for UDT after the age of 5 years has almost certainly an acquired UDT.

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Authors' affiliations

W W M Hack, K Sijstermans, L M van der Voort-Doedens, Department of Paediatrics, Medical Centre Alkmaar, Alkmaar, The Netherlands
J van Dijk, M J Hobbelt-Stoker, M E de Kok, Institution for Youth Health Care, Gemeenschappelijke Gezondheidsdienst Noord-Kennemerland, Alkmaar, The Netherlands

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United Kingdom Paediatric Chronic ITP Registry

We are pleased to announce that the UK Paediatric ITP Registry is now online at www.uk-ityp.org. We would like to encourage UK Paediatricians to participate in this important, prospective national study of childhood immune thrombocytopenic purpura (ITP). The registry has been established for children aged between 2 months and 16 years diagnosed with acute ITP in the past 12 months and will share data with an international registry for ITP (Paediatric and Adult intercontinental Registry (PARC) on chronic ITP). Previous UK audits have established the frequency of acute ITP and bleeding complications in acute ITP but there have been no studies collecting longer term data on those patients who develop chronic ITP.

The primary objective will be to establish the consequences of acute and chronic ITP on the frequency and severity of bleeding symptoms and on the requirement for treatment in these conditions. Secondary objectives include the collection of data on the long-term outcomes of ITP, on the frequency and outcome of intracranial and other significant haemorrhage, and on the various treatment strategies used to increase patients' platelet counts. We anticipate that information collected through the registry will subsequently be used to identify children at high or low risk of significant haemorrhage so that interventional treatment could be offered appropriately.

What work is involved for data collection?

MREC has agreed that all sites participating in this study are exempt from site-specific assessment. There is no need to inform Local Research Ethics Committees (LRECs) about the research. You will need to notify your local R&D department about the research and provide copies of REC application and approval letter, both of which can be downloaded from the website www.uk-ityp.org.

Children with ITP can be registered within 12 months of initial diagnosis. Registration details can be entered directly onto the secure server using any NHS computer. Data requested will consist of standard information stored in the medical notes such as bleeding episodes, therapy required and blood counts. Follow-up information will be requested 6 months after registration: no further information will then be collected on those children whose ITP has resolved whereas data will continue to be collected annually on children with ongoing chronic ITP.

Further information: Dr John Grainger, Consultant Paediatric Haematologist, Royal Manchester Children's Hospital, Manchester, UK; Email john.grainger@cmmc.nhs.uk; Website: www.uk-ityp.org.