Sex and gender issues in competitive sports: investigation of a historical case leads to a new viewpoint

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ABSTRACT
Based on DNA analysis of a historical case, the authors describe how a female athlete can be unknowingly confronted with the consequences of a disorder of sex development resulting in hyperandrogenism emerging early in her sports career. In such a situation, it is harmful and confusing to question sex and gender. Exposure to either a low or high level of endogenous testosterone from puberty is a decisive factor with respect to sexual dimorphism of physical performance. Yet, measurement of testosterone is not the means by which questions of an athlete’s eligibility to compete with either women or men are resolved. The authors discuss that it might be justifiable to use the circulating testosterone level as an endocrinological parameter, to try to arrive at an objective criterion in evaluating what separates women and men in sports competitions, which could prevent the initiation of complicated, lengthy and damaging sex and gender verification procedures.

INTRODUCTION
In 1949, the Dutch track athlete Foeke Dillema (1926–2007) came to prominence on the world athletic stage. She started to rival Fanny Blankers-Koen, the world-famous Dutch track athlete who won four gold medals during the 1948 Summer Olympics in London and was elected Female Athlete of the Century by the International Association of Athletics Federations (IAAF) in 1999. In contrast, Dillema’s career was of short duration, with a dramatic ending. In 1950, she was expelled for life by the Royal Dutch Athletics Federation, due to the results of a ‘sex test’, for which details or results were never revealed and no records are available. Her 1950 national record of 24.1 s for the 200 m, which she took from Fanny Blankers-Koen, was erased, and only after her death 57 years later was she reinstated by the Royal Dutch Athletics Federation (figure 1).1

The verification of the sex of athletes has been an issue for many decades. It should be noted that reports and reviews on this topic refer to gender verification, rather than sex verification.2–4 However, what counts in competitive sports is a person’s sex characteristics. Trying to avoid the word sex, given its charged nature, can only cause confusion.5,6 Herein, we will use the term sex for the biological and physiological characteristics that define men and women, as compared to gender and gender identity in reference to the socially and individually perceived sexual identity of an individual from birth to puberty and adulthood.5,7,8

Sex verification in 1950 was based solely on physical examination, predating hormone assays or sex chromosome analysis. Following discovery of the Barr body in female cells in 1949, it took some 12 years before it was known that this body represents an inactivated X chromosome; from the late 1960s its detection was used in sex verification tests in the context of sports competitions.2–4 Subsequent tests focused on the male-specific region of the Y chromosome, particularly the male sex determining SRY gene.10 However, opposition to sex verification for female athletes with laboratory-based genetic testing developed in the 1970s and 1980s, because these tests did not encompass the complexities of disorders of sex development (DSDs). Since the 2000 Summer Olympics, questioned sex and gender is evaluated on a case-by-case basis by a team of specialists in the areas of endocrinology, genetics, gynaecology and psychology.3–5

To broaden the perspective on sportswomen confronted with questioned sex characteristics, we have investigated the case of Foeke Dillema, with informed consent from her heirs, by means of DNA analysis of samples from worn clothing. Appreciating the nature of the samples tested, we applied DNA methodology and lab quality standards used in human forensics. Our DNA analysis indicates that Foeke Dillema had a 46,XX/46,XY mosaic condition with a rare origin, which we interpret as leading to hyperandrogenism from her puberty. Based on this historical case we discuss that, if a sportswoman is confronted with signs of a DSD early in her sports career, it is harmful and confusing to question such a person’s sex and gender. Rather, we suggest that it is necessary to try to arrive at an objective criterion in evaluating what separates women and men in sports competitions.

RECONSTRUCTION OF A HISTORICAL CASE
From the combined genotyping and DNA quantification results, we conclude that Foeke Dillema was a 46,XX/46,XY mosaic, with equal numbers of both genetic cell types at least in her skin (online supplementary data). In the fetal gonads of a 46,XX/46,XY mosaic, the tissue ratio of XX:XY cells will push the bipotential gonads to become either ovaries or testes, or both. A preponderance of 46,XX cells in fetal gonads can
ment. From all available data, including the present DNA evi-
dence, we deduce that Dillema had an ovotesticular DSD with
a predominance of ovarian tissue. At the onset of puberty,
gonadotropic stimulation of the gonads most likely led to
marked activation of steroidogenesis – the production of both

Figure 1  Foekje Dillema (in white shirt on the left) together with Fanny Blankers-Koen, on the Olympic Day, 18 June 1950, in the Olympic Stadium, Amsterdam, when 60,000 spectators witnessed Dillema winning the 200 m in 24.1 s, in a race in which Blankers-Koen did not participate.1 Photo: Ben van Meerendonk (collection International Institute of Social History, Amsterdam).

The X:Y ratio in the adult skin, as we obtained from
Dillema’s clothes, does not provide any information about the
ratio in her fetal gonads. However, Foekje Dillema was for-
mally registered as a female at birth, and she was raised as a
girl, according to all accounts, including those available from
her family. Hence, she is unlikely to have been exposed to a
markedly elevated level of testosterone during fetal develop-
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le to the development of ovaries, but with some 46,XY
testis tissue present in one or both of these ovaries. Such an
ovotestis condition, which can also occur in the form of a
complete ovary and a complete testis on either side, has been
referred to with the term true hermaphroditism.11,12 The most
common karyotype found in true hermaphroditism is 46,XX,
followed by 46,XX/46,XY chimerism and mosaicism.12 We
would like to emphasise that the term hermaphrodite, as
well as other terms such as intersex, need to be replaced by
the DSDs classification proposed by consensus in 2006,13 for
the simple reason that the terms hermaphrodite and inter-
sex cannot, and should not, be applied to human individuals.
Instead, a true hermaphrodite is correctly referred to as either
a female or male individual with ovotesticular DSD.13 When
ovotestis formation is confined to only one of the gonads, the
other gonad and the ovarian part of the ovotestis can func-
tion as steroidogenic ovarian tissue.11,12 The testicular part
of the fetal ovotestis will produce anti-Müllerian hormone,
insulin-like factor 3 and testosterone,14 acting towards regres-
sion of Müllerian ducts, initiation of testis descent and partial
virilisation by circulating androgens, respectively, but none of
these effects might reach a level where overt fetal virilisation
leads to the birth of a boy. Hence, an individual with ovotes-
ticular DSD often is raised as a girl,12 and also experiences a
female gender identity (described below).

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SEX CHARACTERISTICS, GENDER IDENTITY AND
TESTOSTERONE

To have, or not to have, a Y chromosome is the primary deci-
sive factor in human sexual differentiation, but there are exceptions. A prominent example is offered by 46,XY females
who have the complete form of androgen insensitivity syn-
drome (cAIS), when the testes produce testosterone but the
body is not able to respond to androgens (testosterone and its
more powerful metabolite dihydrotestosterone) due to muta-
tion of the X-encoded androgen receptor.14 Consequently, these individuals are born and raised as girls, and have a
female gender identity.15,16 Action of testosterone through
binding to the androgen receptor in the developing fetal brain
is the predominant factor in programming human male gen-
der identity,7,17 and the female gender identity of 46,XY cAIS
women is explained by loss of this androgenic effect. In sports,
46,XY cAIS women can be expected to have a disadvantage
compared to 46,XX women with a functional androgen recep-
tor, the latter profiting from stimulation of muscle strength by
a low level of circulating testosterone.18,19

The biological basis for sex segregation in sports is the con-
sequence of long-term endogenous androgen exposure of men
after puberty.20 It cannot be excluded that proteins encoded by
genes in the male-specific region of the Y chromosome (MSY)21
might act together with androgens, widening the physiologi-
cal gap between women and men. However, such a role for
MSY genes will be minor, compared to the predominant role
of androgen action. In men, the postpubertal testosterone level
is a proven dose-dependent factor when muscle strength and
other physiological factors such as the blood haemoglobin level
come into play.22 A moderate pubertal and postpubertal excess
of testosterone in a young woman can give extra muscle devel-
opment and other signs of hyperandrogenism, but it would be
a rude error to even suggest that this would affect her female
gender identity.

Competitive athletes exploit fortunate combinations of
natural differences in physical and mental personal charac-
teristics, including individual variation of the endogenous
testosterone level. The World Anti-Doping Agency states that
an athlete’s sample will be found positive if the concen-
tration of an endogenous androgenic steroid hormone is
above the range normally found in humans, and is not likely
consistent with normal endogenous production, unless the
elevated concentration of the steroid hormone (or metabo-
lites or markers) is attributable to a physiological or patholog-
ical condition.23 Strictly speaking, a female athlete is free to
benefit from any endogenous source of androgen production.
Some female athletes may benefit, probably to a small extent,
from increased androgen production originating from a poly-
cystic ovary.19 This is viewed as acceptable by the IAAF, who
stated that conditions that may provide some advantages but
nevertheless are acceptable include congenital adrenal hyper-
plasia, androgen-producing tumours and an ovariary andro-
gen excess associated with a polycystic ovary.24 According
to these regulations, hyperandrogenism caused by ovotes-
ticular DSD would be unacceptable only if sex and gender

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Doping controls fighting against the use of exogenous androgens, binding proteins, etc. 25 – 27 Normative ranges have variations in measurement, metabolites, circadian and other endogenous testosterone level will require a detailed discussion. Re-entry, leaving no trace of a sex and gender discussion. One level would have been detected, possible treatment to be ethically justifiable, given the fact that it would be in the individual’s own interest to prevent symptoms of long-term hyperandrogenism. The causes and consequences of a high testosterone level can be dealt with in private, not in public, and, most importantly, without questioning gender identity. Hence, there is a need to reconsider the situation.

It might be considered to set an upper limit for the circulating total testosterone level for sportswomen. In cases of a AIS, a high testosterone level would be of no significance. In any other case where the total testosterone level is found to exceed a set limit, causes and consequences need to be resolved before the sportswoman (re-)enters sports competition. This would be ethically justifiable, given the fact that it would be in the individual’s own interest to prevent symptoms of long-term hyperandrogenism. The causes and consequences of a high-testosterone level can be dealt with in private, not in public, and, most importantly, without questioning gender identity. The eligibility of Dillema to compete might still be questioned. Rather, there is a need for an objective and relevant criterion in evaluating what separates women and men in sports competitions.

What this study adds

Describing a historical case, this study puts forward the notion that societal appreciation of sex and gender issues in highly competitive sports requires discussion and understanding of relevant biomedical knowledge. However, the authenticity of an adult individual’s sex and gender identity should not be questioned. Rather, there is a need for an objective and relevant criterion in evaluating what separates women and men in sports competitions.

verification would provide evidence that the female athlete in fact is a man. However, we consider it highly unlikely that any individual would aim to participate in sports competitions in conflict with his or her gender identity. There is no problem in sports at large that warrants an examination, initiated by a sports federation, of the authenticity of an adult individual’s sex and gender. Hence, there is a need to reconsider the situation.

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The historical case described herein concerns ovotesticular DSD, where the amount of steroidogenic testicular tissue will determine if the affected person develops as a woman or as a man, regarding both gender and sexual characteristics. As such, this type of DSD can be viewed as a paradigm, demonstrating that the testosterone level might offer an objective parameter to separate the sexes, if required. In fact, this parameter is already implemented, in the context of sports. Athletes (46,XY and androgen sensitive) who have undergone male-to-female sex reassignment are welcome to engage in sports competitions from 2 years after the sex change, as of the Olympic Games 2004 in Athens, according to fortunate and emancipative regulations by the International Olympic Committee. 38 Similarly, athletes (46,XX and androgen sensitive) who have undergone female-to-male sex reassignment can compete, but they will receive exogenous testosterone. For female-to-male sex-reassigned individuals receiving testosterone supplementation, a total testosterone level of around 30 nmol/l has been reported. 39 Perhaps, one day we may witness a talented 46,XX sex-reassigned male who is able to successfully compete with 46,XY males, thanks also to approved testosterone supplementation. Men exposed to stress and exhaustion face difficulty in maintaining their endogenous testosterone level, 40 – 42 which might imply an advantage for 46,XX sex-reassigned males, particularly in endurance sports. Similarly, a therapeutic use exemption for long-term testosterone administration in 46,XY men, to compensate for a secondary loss of gonadal testosterone production, might provide an advantage. The above serves to illustrate the point that current concepts and regulations regarding the relationship between sex and testosterone in sports offer room for consideration. We feel that this should be taken as a starting point to discuss the circulating testosterone level as a relevant criterion in evaluating what separates women and men in sports competitions.

The present report is not meant to provide a guideline, which would require detailed analysis of total testosterone levels in large numbers of female and male athletes in relation to possible confounding factors, and consensus meetings. Rather, we aim to contribute to an open discussion involving experts from the fields of biology, medicine, genetics, psychology, sports and ethics, to accomplish a procedure which would respect the authenticity of an adult individual’s sex and gender identity.
Note: On 12 April 2011, the International Association of Athletics Federations (IAAF) announced the adoption of new rules and regulations governing the eligibility of females with hyperandrogenism to participate in women’s competition, which will come into force from 1 May 2011 (http://www.iaaf.org/aboutiaaf/news/newsid=59746.html).

It appears that these new IAAF rules, as announced, are in full agreement with the viewpoint expressed in our article, which at the time of the IAAF announcement was already in press with the British Journal of Sports Medicine. We would like to emphasise that our viewpoint was composed independently from IAAF, and that none of the authors has been in contact with the respective IAAF expert working group.

Acknowledgements The authors are very grateful to the heirs of Foekje Dillenma for their frank and considerate willingness to dedicate information and materials to the present historical perspective on female athletes confronted with questioned gender issues. For the photograph of figure 1 we obtained right of use from the International Institute of Social History, Amsterdam.

Funding KNB and MK were supported by the Netherlands Forensic Institute (NFI), and the Netherlands Genomics Initiative (NGI)/Netherlands Organization for Scientific Research (NWO) within the framework of the Forensic Genomics Consortium Netherlands (FGCN).

Patient consent Obtained.

Contributors KNB and MK designed the experiments, KNB carried out the experiments, MK provided lab equipment and test materials, all authors contributed to the data interpretation, the writing of this manuscript was led by JAG, and all authors approved the final manuscript.

Provenance and peer review Not commissioned; externally peer reviewed.

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Br J Sports Med  published online May 3, 2011

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