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POLYORCHIDISM AND FERTILITY

POLIORHIDIZAM I FERTILITET

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Ključne reči
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Abstract

Introduction & Objective: Polyorchidism is a rare congenital anomaly. In this study we add two more cases to the description of Polyorchidism in a literature.
Material & Methods: In this article two patients at 17 and 19 years old with Polyorchidim were described. We carried out their fertilizing ability with following methods: medical history and physical examination, spermatological analysis of the ejaculate according WHO recommendation, light- and transmission electron microscopy of the testicular tissue and spermatozoa, hormonal analysis of LH, FSH and Testosterone plasma blood concentrations by conventional methods.
Results: In cases with supplemental third testis, the histological picture evidenced of full spermatogenesis, represented by all cells spermatogenic line. The spermatological studies showed that the physical characteristics of seminal plasma and quantitative changes in spermatogenesis were within the normal parameters. In this cases 60% of the sperm was with rapidly progressive movement, i.e. highest rating of motility. We proved the presence of sperm with normal and abnormal configuration forms in 51 and 49%, respectively. The hormonal assays on patients with Polyorchidism demonstrated the presence of preserved endocrine function
Conclusion: Polyorchidism is the only inherited male reproductive tract disease in which man is capable of fathering. This is the first comprehensive study on the preserved fertility of Polyorchidism patients.

INTRODUCTION

Polyorchidism is an uncommon congenital anomaly [(1)]. The occurrence of Polyorchidism (presence of more than two testis confirmed by histology) most probably attributable to the abnormal embryonic development of gonads - dividing the gonad before the 8th gestation period [(2, 3)] and in a more cases the most supernumerary testes (64%) were drained by a vas deferens.

The first observation of this anomaly was of Lane in 1935 [(4)]. It is a rare malphormation of male reproductive tract [(5)]. Median patient age at detection was 17 years [(1)], and according Spranger et al.[(2)] approximately 50% of cases are seen between 15 and 25 years old.

Triorchidism was the most common type [(6)]. In the literature describes the cases of the presence of 4 gonads [(1)].

About 75% of supernumerary testes are intrascrotal, with another 20% located in the inquinal canal and 5% in the retroperitoneal space [(7)]. Left side [(1, 8)] demonstrated predominance (64.5% of the cases).

In their meta-analysis of literature, Bergholz and Wenke [(1)] described that most cases were found during surgery for other symptoms including inguinal hernia, undescended testicle, hydrocele [(9)], testicular torsion [(10)] and scrotal pain. Very often the malformation may be associated with pathology of the epidydimis or ductus deferens [(11, 12)]. Only 16% of patients complained of an accessory mass without any symptoms. Neoplasms were found in 9 cases (6.4%), of which 8 were malignant and 1 was benign. Data available for 7 malignomas (88%) revealed that all were in cryptorchid supernumerary testes.

Polyorchidism is the only inherited male reproductive tract disease in which man is capable of fathering. Typical in this malformation of the testis is that there is preserved spermatogenesis [(1, 13, 14)], and basal gonadotropine and androgen hormones concentrations are in a normal value [(9, 14, 15)].

In this study we add two more cases to the description of Polyorchidism in a literature.

MATERIALS AND METHODS

In this article two patients at 17 and 19 years old with Polyorchidism were described.

We carried out investigations to determine their fertilizing ability by the following methods:

● *Medical history and physical examination;*

● *Spermatological analysis of the ejaculate* according to WHO (16) recommendation;

● *Light microscopy* of the testicular tissue and spermatozoa after routine staining with Yashkovski, Papanicolaou, hematoxyline-eosin, etc. Light microscope studies were carried out on stained tissue/ejaculate smears. Hundred spermatozoa were counted and measured in each smear using the following magnifications – 10X40 and X00. The percentage of spermatozoa with normal and altered head was calculated for each sample. Tissue and spermatozoa were studied and counted on a light microscope Carl Zeiss, Jena, Germany;

● *Transmission electron microscopy* /„Opton" EM 109/ for evaluation of ultrastructural changes in the sperm cells;

● *Hormonal analysis of LH, FSH and Testosterone* plasma blood concentrations by conventional methods using commercial kits (Immunotech, France).

● *Definition of rating motility:*

The percentage of sperm with rapid progressive movement

(Nº 1 form of movement)

The percentage of sperm with slowly progressive movement

(from Nº 2 to Nº 4 forms of movement)

Motility without progression

(from Nº 5 to Nº 10 forms of movement)

The percentage of immotile sperm

(Nº 11 form of movement

Forms of movement: *Nº1 - rapidly progressive movement; Nº2 - circular vibratory movement; Nº3 - to throw movement; Nº4 - rotational movement; Nº5 - asymmetric head and/or tail; Nº6 - cytoplasmic sperm with tail; Nº7 - sperm with agglutination; Nº8 - circular, oscillating, vibrating movement; Nº9 – wavy movement; Nº10 – whip movement; Nº11 - fixed sperm.*

● *The following criteria for evaluation of the fertilizing ability in Polyorchidism patients were used:*

In cases of polyorchidism with supplementary third testis, the histological picture demonstrated full spermatogenesis, represented by all spermatogenic line cells (Fig. 1A, B).

At the ultrastructural level, spermatogonia and spermatocytes with relatively well preserved morphology were observed. Many vacuoles were seen in the cytoplasm of some spermatocytes (Fig. 1C), while chromatin fragmentation in the nucleus with preserved sinaptonemal complexes is found in other spermatocytes (Fig. 1D). Contacts between some spermatocytes and the neighboring Sertoli cells were disrupted. There were tears of Chris in mitochondria (see Fig. 1D).

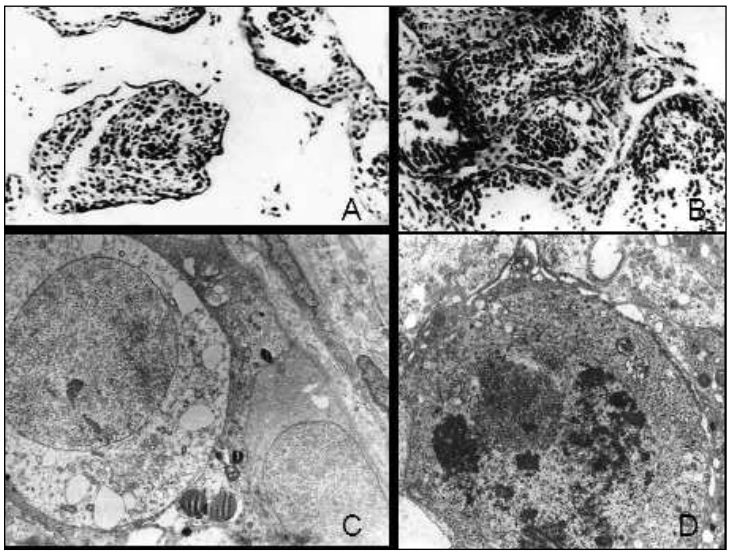

Figure 1. *(A) Histological picture of the right testis and (B) accessory right testis in Polyorchidism. HE x 100; (C) Spermatocytes from a Polyorchidism patient with relatively well preserved morphology. X 7000; (D) Spermatocyte cells with damaged structure and contacts with neighboring cells. X 12000*

The conducted spermatological studies showed that the physical characteristics of seminal plasma were within the normal parameters. The ejaculate volume in Polyorchidism (2.81 0.02 ml) was in the range of physiological values (3.5 0.31 ml). The pH values (7.5) were within physiological limits (7.62) in the normal function of accessory glands. There was a normal viscosity without agglutination.

Quantitative changes in spermatogenesis in this pathology were not found out (sperm count – 72 mil/ml).

Along with concentration, the gamete motility was an important link in the fertilizing ability. Speed was an integral part of the motility, which was usually directly proportional to the sperm movement (Table 1, 2).

Preserved fertilizing ability
(normospermia + ≥60% motility + ≥10 μ/sec velocity + 70% ♂ with normal morphology)

Relatively preserved fertilizing ability
(normospermia +≥60% motility + ≤10 μ/sec velocity + ≤ 70% ♂ with normal morphology)
(oligozoospermia Gr. I + ≥ 60% motility + ≥10 μ/sec velocity + ≥ 70% ♂ with normal morphology)

Poor fertilizing ability
(oligozoospermia Gr. I + ≤ 60% motility + ≤10 μ/sec velocity + ≤ 70% ♂ with normal morphology)
(oligozoospermia Gr. II-III + ≤ 60% motility + ≤10 μ/sec velocity + ≤70% ♂ with normal morphology)

Missing fertilizing ability
(azoospermia + aspermia)

	Motility (%)			Velosity (μ/sec)
	Progressive motility	Rapid motility	Immotile	
Polyorchidism /n=2/	60.00± 0.36	13.00 ± 1.33	27.00± 0.81	12.00± 1.22
Control group /n=30/	63.89 ± 5.05	15.13 ± 1.67	20.98 ± 4.4	16.31 ± 1.38

Table 1. *Sperm motility and velocity in cases of Polyorchidism*

	FORMS OF MOVEMENT (%)										
	N^{o} 1	N^{o} 2	N^{o} 3	N^{o} 4	N^{o} 5	N^{o} 6	N^{o} 7	N^{o} 8	N^{o} 9	N^{o} 10	N^{o} 11
POLYORCHIDISM	60	9	–	4	–	–	–	7	–	–	20
CONTROL GROUP	65	7	–	2	–	1	–	5	–	–	20

Table 2. *Forms of sperm movement*

Forms of movement: *N^{o}1 - rapidly progressive movement; N^{o}2 - circular vibratory movement; N^{o}3 - to throw movement; N^{o}4 - rotational movement; N^{o}5 - asymmetric head and/or tail; N^{o}6 - cytoplasmic sperm with tail; N^{o}7 - sperm with agglutination; N^{o}8 - circular, oscillating, vibrating movement; N^{o}9 – wavy movement; N^{o}10 – whip movement;N^{o}11 - fixed sperm.*

In assessing sperm motility useful data on the rating of motility can be extracted. In this Polyorchidism case (see Tabl. 2), 60% of the sperm was with rapidly progressive movement, i.e. highest rating of motility. The percentage of sperm with slowly progressive movement was 13. Motility without progression and immotile sperm were reported in 7 and 20%, respectively, which were with the lowest score (27%).

Based on our morphological studies, we proved the presence of sperm with normal and abnormal configuration forms in 51 and 49%, respectively. Below in the Table 3 are presented the results obtained by light-microscopic studies of abnormal sperm. The highest percentage of gamete changes was found in the head structure (23%) and flagellum (17%). Mixed sperm enjured configuration were proved in 7%.

The hormonal assays on patients with Polyorchidism demonstrated the presence of preserved endocrine function. The value of gonadotropine hormones LH (8.43 mUI/ml), FSH (5.97 mUI/ml), and the steroid hormone Testosterone (2.14 ± 0.01 ng/ml) were in the normal range of the basal plasma concentrations (8.99 ± 2.02 mUI/ml, 6.90 ± 1.12 mUI/ml and 3.02 ± 0.41 ng/ml, respectively).

DISCUSSION

As we have already mentioned, Polyorchidism is the only congenital disease of the male reproductive tract in which man is capable of fathering [1]. This is confirmed by our results.

This malphormation is accompanied by fully preserved fertility with a normal sperm number, motility and velocity. Similar data are reported by other authors [1, 14].

The preserved spermatogenesis demonstrated at the light- and electron-microscopic level, as well as the finding of Pomara et al. [13], observed in both testes revealed normal spermatogonial proliferation and normal meiotic progression up to the second spermatocyte stage. Many spermatids showed clear piknotic degeneration, and the base membrane was lightly thickened. There is also preserved endocrine regulation of the pituitary-gonadal axis impact on the reproductive process. The basal level of gonadotropic and androgenic hormones was within the normal values [14, 15].

To conclude, this is the first comprehensive study on the preserved fertility of Polyorchidism patients.

CONCLUSION

Polyorchidism is the only inherited male reproductive tract disease in which man is capable of fathering. This is the first comprehensive study on the preserved fertility of Polyorchidism patients.

Conflict of Interest: None of the contributing authors have any conflict of interest, including specific financial interests or relationships and affiliations relevant to the subject matter or materials discussed in the manuscript. In lieu of a formal ethics committee, the principles of the Helsinki Declaration were followed. All human subjects provided written informed consent with guarantees of confidentiality.

Table 3. *Spermatozoa with injured configuration (%) in cases with Polyorchidism*

I. Sperm head anomalies		**29**
	1. Macrocephalic head	**7**
	2. Microcephalic head	**6**
	With normal form	2
	With spherical form	4
	3. Round head	**4**
	With flagellum	3
	No flagellum	1
	4. Elongate haed	**11**
	With flagellum	4
	No flagellum	7
	5. Two head	**1**
II. Sperm flagellum anomalies		21
	1. Coiled tail	**4**
	2. Short tail	**9**
	3. Two tail	**3**
	4. No head	**1**
III. Cytoplasmic droplet		**2**
	1. around head	**1**
	2. around neck	**1**
IV. Mixed anomalies		**7**
	1. Macrocephalic head and coiled tail	**2**
	2. Round head and short tail	**1**
	3. Round head and cytoplasmic droplet	**1**
	4. Elongate head and coiled tail	**2**
	5. Elongate head and bent tail	**1**
♂ **%**	**with injured configuration**	**49**

Sažetak

Uvod i cilj: Poliorhidizam je retka kongenitalna anomalija. Ovim radom dodali smo još dva slučaja opisivanju poliorhidizma u literaturi.
Materijal i metode: U ovom članku su opisana dva pacijenta, od 17 i 19 godina, sa poliorhidizmom. Ispitali smo njihovu sposobnost fertilizacije primenom sledećih metoda: istorija bolesti i fizičko ispitivanje, spermatološka analiza ejakulata prema preporukama WHO, svetlosna i transmisiona elektrononska mikroskopija tkiva testisa i spermatozoida, hormonalna analiza LH, FSH i koncentracija testosterona u krvnoj plazmi konvencionalnim metodama.
Rezultati: U slučajevima sa prisustvom trećeg testisa, histološka slika pokazuje potpunu spermatogenezu i prisustvo svih ćelija spermatogene linije. Spermatološka ispitivanja pokazuju da su fizičke osobine semene plazme i kvantitativne promene u spermatogenezi bile u okviru normalnih parametara. U ovim slučajevima 60% sperme je bilo sa brzim, progresivnim kretanjem, odnosno s najvećom pokretljivošću. Dokazali smo prisustvo sperme s normalnom i abnormalnom konfiguracijom u 51% i 49%, redom. Hormonski test na pacijentima sa poliorhidizmom pokazao je prisustvo sačuvane endokrine funkcije.
Zaključak: Polioridizam je jedino nasledno oboljenje reproduktivnog trakta muškaraca kod kojeg je muškarac sposoban da ostavi potomstvo. Ovo je prvo sveobuhvatno ispitivanje o sačuvanom fertilitetu pacijenata sa poliorhidizmom.

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