Persistent Urogenital Membrane in Female Children Over the Vaginal Vestibule

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THE Urogenital membrane is the cranial part of the cloacal membrane after the separation of the urogenital sinus from the rectum by the Urorectal septum. The caudal end of the cloacal membrane is known as anal membrane covering the anal canal in the developing foetus. The anal membrane completely ruptures and provides the anal opening except in few cases it remains as such and results into the condition called as imperforate anus. The similar anamoly may be present in urogenital membrane over the vestibular area by its persistance in female children. Here we wish to present in this article such seven female children, who had persistant membrane over the vestibular area except a small opening for urinary passage, giving anxiety to their parents.

Embryology

The hind gut opens into the cloaca which is covered by the cloacal membrane. It is divided into two by the urorectal septum. The anterior space is called as urogenital sinus and posterior is rectum and anal canal. The anterior part of cloacal membrane covering the urogenital sinus is labelled as urogenital membrane and posteriorly its part covering the anal canal opening is called as anal membrane. In the female foetus, the urogenital sinus becomes shallow and urethra is formed anteriorly from this. The vagina opens in its posterior wall, from above. The both openings i.e. urethral and vaginal opening open in that shallow space of urogenital sinus, which is known as vestibule later on. The vestibule is surrounded by labia minora from either sides developing from genital folds, and anteriorly clitoris developing from genital tubercule, is situated. The posterior end of labia minora fuses and forms fourchette. There is a few milimeter wide triangular skin ledge creating a fossa underneath the fourchette known as vestibular fossa which never extends anteriorly upto the clitoris in normal females. But the membrane may remain intact over its vestibule on the rare occasion except a small opening for the urinary passage.

Observation

Seven female children were brought

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to us with the similar problem of having intact vestibular membrane. The children were in betv een 1 month and $2\frac{1}{2}$ years age group right from birth as shown in the table 1. The chronological position of the child among her brother sister is as shown in table 1. In all cases the abnormality was detected either by mother, or attending Ayah or by personal Doctor, but mostly by the mother. The moment this condition was noticed, the parents became too much worried about the fate of the child, especially from the reproduction point of view at the puberty. Two parents started doubting about the sex of the child. Most of the cases were asymptomatic except in

Table 1

Sr. No.	Age	Position in Sibling	Antenatal	Symptoms/Signs	Associated abnormality	Treatment
1	6 months	4th	FTND	Complete membrane +nt., Small Opening +nt., Bulging during micturation.	Nil	Blunt Probe separation.
2	2 months	3rd	FTND, X-ray Pelvis of mother for suspected breach.	Complete membrane +nt., Small opening +nt., Asymptomatic.	Syndactyly Rt. Hand	Blunt probe separation.
3	8 months	1st	FTND, Home Delivery.	Complete membrane +nt., Small Opening +nt., Bulging during micturation.	Nil	Sharp Scissors separation.
4	1 month	1st	FTND, Broncho dilators to mother for Br. Asthama.	Complete membrane +nt., Small Opening, Asymptomatic.	Nil	Blunt Probe Separation.
5	5 months	3rd	FTND.	Complete membrane +nt., Small Opening +nt., Dysuria.	Nil	Blunt Probe separation.
6	1 Year	2nd	FTND, Home Delivery	Complete membrane +nt.. Small Opening +nt., Asymptomatic.	Systolic murmur	Sharp Scissor separation.
7	$2\frac{1}{2}$ Years	1st	FTND.	Partial membrane +nt., Two openings, Dysuria.	Nil	Blunt probe separation.

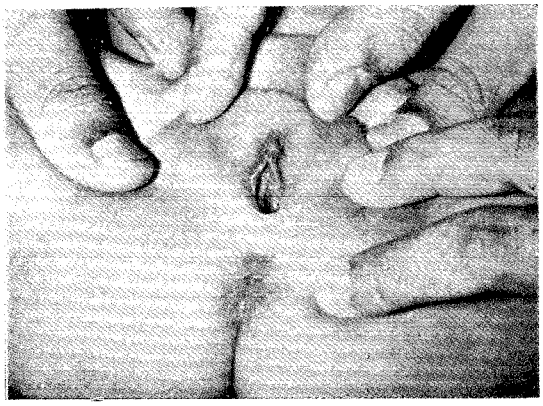
F. T. N. D. = Full Term Normal Delivery. + nt. = Present

four cases. Two had dysurea and two had bulging of the membrane during micturation. There was no history suggestive of any infection in the genitalia. In one case who had dysurea, had another small opening at the fourchette, consulted some one and was subjected for urethral dilatation without breaking the membrane and she was directed to us for the final treatment.

The detail antenatal and natal history alongwith their family history, was noted. But no definite positive clue could be traced in any of them. All were delivered at their full term, five at maternity hospital and two in their homes with the help of midwife. Only one father had polydactyly in left hand. Mostly the children were belonging to middle class and poor class families, socio-economically.

On physical examination, apparantly only one child had syndactyly of middle and ring fingers of the right hand, rest were quite normal. Once their cardiovascular system was examined, one child had doubtful systolic murmur in mitral area, which underwent thorough cardiovascular checkup by physician without any definite finding in his heart. Remaining system of the body were found in sound states.

The careful examination of external genital was performed. The anal opening were found in their normal situation, with their good sphincteric control. The pubic symphasis could be felt in normal place well approximated. The clitoris, with its hood was of average size and shape, having very small opening, on its under surface, through which child was passing urine. Except this small opening there was complete covering over the vestibule by a thin membrane (fig. 1), being



Figi 1—Pre-operative Photograph showing complete covering of vestibule and small opening on the under surface of clitoris.

exception only in one case who had another opening in the fourchette area. The labia majora folds could be marked out, but both the minora lips were forming the continuity of the membrane, and one was unable to mark out them separately. There was no sign of old inflammation in form of scarring etc. A blunt probe could be passed through the small opening, underneath the membrane. It was passed gently, in horizontal direction posteriorly, to avoid trauma to the urethra and vagina, and could be negotiated till the fourchette, confirming the presence of vestibular space, covered by its membrane. In case who had two openings, the probe could be passed through and through. The treatment in these children was very simple, which could be carried out in the out patient department, without the need of hospitalization of patients and without giving any anaesthesia to them. The time required was

very minimal and without any post-operative complication with a very easy technique as follows :

A metallic probe was passed gently keeping it in the middle line, in horizontal direction till the posterior end of the membrane (Fig. 2). The membrane got separated with mild, downwards and outward force towards the anal side. There was hardly any bleeding from the separated edges. On opening of the membrane (Fig. 3) the normal urethral and vaginal openings were found in the vestibule and labia minora could be marked out. But in two cases this blunt procedure did not help, so they were subjected for separation of the membrane by sharp scissors. The cut edges were touched with tincture benzoin co. to stop the mild oozing. There was no need of any surgical or manual interference to keep the separated edges apart. The parents were advised to apply neosporine ointment at the edges for a week. So far, in our 3 months to 3 years follow up, none of our cases needed reseparation for its recurrence.

Discussion

This condition resembling clinically, is described as fused labia in the literature. According to them, the fusion takes place due to oestrogen deficiency or due to the excess of androgen activity in the body in these children. The other groups think it, as result of local inflammation, resulting in the labial adhesion. But in our cases we were unable to find any clue of old inflammation locally and this condition can be explained on the embryological basis to be intact urogenital membrane in the vestibular area as in cases of imperforate anus. Moreover there was no sign of excessive andogenic activity in the form of enlarged

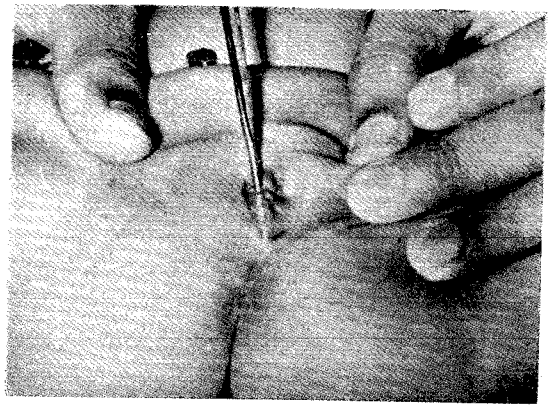
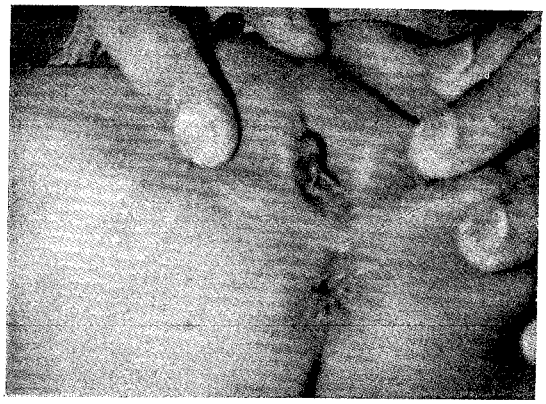


Fig. 2—Pre-operative photograph with probe under the membrane.



Fiu. 3—Post-operative photograph after separation of the membrane.

clitoris, or so, thus according to them vestibular membrane has already ruptured and again fusion of the two labial folds has taken place, sounds unlikely without any definite local findings.

There is controversy regarding its treatment also. This condition is usually harmless and resolves spontaneously at puberty (Anderson 1956). But they should be

treated to correct the urinary problem and to relieve fhe anxiety of the parents. Several authors have recently advised topical oestrogen therapy (Hoffman 1968, Althek 1972) with its uncertain results and requires prolong therapy atleast from 3 weeks to months and in some cases it does not work well then ultimately surgical interference has to be done. Moreover, in some cases local complications in form of vulval pigmentation and erythema and in some cases systemic side effects have also been noticed while giving prolonged therapy in form of oestrogen cream or ointment locally or systemic therapy of oestrogen to cover up its difficiency respectively. In our opinion due to non-availability of sophisticated laboratory help, & to avoid prolonged therapy with uncertainty of its results, the surgical separation of the membrane is the best method to be adopted, which gives hardly any problem to the child or to their parents during the procedure or later on. We have not encountered the recurrence so far in our follow up as reported by Nawlia et al (1949), Caparaw and Greenberg (1972) in their surgically treated cases. Therefore we think that this clinical condition is a congenital developmental abnormality in which vestibular membrane, a part of urogenital membrane has not ruptured during gestation and persisted as such as in cases of imperforate anus with persistant anal membrane. The surgical separation either by blunt probe or with the help of sharp scissor is the best method of their management.

Summary

This article consists of seven female children with alike congenital problem creating great psychological set back to their parents. The children had persistant urogenital membrane in vestibular area except with a very small opening for urinary passage. This being a rare congenital anomaly with its very simple technique of its cure, to relieve the mental worry of the parents, prompted us to put this in the literature for further exploration of its etiology and existence.

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