

PNEUMATURIA

Report of a Case in a Diabetic With Review of the Literature

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The passage by the urethra of urine containing gas or air is known as pneumaturia. Senator,¹ in 1891, suggested a classification of this condition into three groups, as follows: 1, pneumaturia resulting from air introduced into the bladder by means of instrumentation or catheterization; 2, pneumaturia resulting from gas entering the bladder by way of a fistulous connection, e.g. vesicovaginal fistula; and 3, pneumaturia resulting from gas produced by decomposition or fermentation of urine in the bladder. Group 3 may be further subdivided into the cases with and without glycosuria. This paper is concerned with cases falling into the category of the third group, with special emphasis on the association with glycosuria.

We have reviewed the literature since the first report in 1860 and have collected 20 cases of the primary type of pneumaturia with glycosuria (Table 1), and 19 without glycosuria (Table 2). In five of the cases with glycosuria, the diagnosis of true diabetes is uncertain. In one of these, pneumaturia and glycosuria observed during a state of coma later disappeared.² In another case, there was only a past history of glycosuria prior to the onset of pneumaturia.³ In the remaining three cases,^{4,5} glycosuria was not present at the onset of the pneumaturia, but glucose was later found in the urine intermittently. As for the nonglycosuric cases, two of the four patients described by Adrian and Hamm⁶ may have had vesico-enteric fistulae secondary to carcinoma, although definite information was not given.

The first mention of what is today considered pneu-

maturia was in the "Curiosities of Nature," published in 1671.⁷ A man was reported to have passed wind by the urethra with or without the passage of urine. Ribes,⁸ when professor to the Medical School of Paris, showed his students an infant whose only malady was the passage of gas by way of the penis. The urine was found to be normal. Celui de Poirry⁹ in 1820 stated that the bladder can contain air under certain circumstances.

It was Boismont,¹⁰ however, who in 1825 was the first to mention the spontaneous production of gas in the urinary bladder. Roche¹¹ in 1836 reaffirmed this. Chomel¹² added the suggestion that the gas may be due to fermentation.

Up to 1860 all the cases reported were instances of fistulous communication between the bowel and the bladder. In that year Raciborski¹³ described in detail the first case in which no fistulous connection between the bladder and any other organ was found. He named the condition pneumo-uria, because his patient had developed the gas spontaneously in the bladder. The gas was odorless, but Raciborski had seen some cases in which the gas had the odor of hydrogen sulfide.

Keyes¹⁴ in 1882 reported the first cases in the American literature. They were the result of instrumentation in two men with prostatic hypertrophy. Guiard¹⁵ first pointed out the occurrence of pneumaturia in diabetes in 1883 and reported four cases. A series of case reports appeared since then.¹⁶⁻³⁶

Mueller¹⁸ reported the first analysis of the gas found in diabetic pneumaturia; it contained a predominance

TABLE 1 GLYCOSURIC CASES REPORTED IN THE LITERATURE

Author	Year	Age and Sex	Organism	Pathology
Guiard	1883	77 M	?	Bladder stone
		60 M	?	Cystitis; suppurative pyelonephritis
		51 M	?	Bladder stone
		60 M	?	Enlarged prostate
Dumenil	1883	45 M	?	Urethral stricture
Thomas	1885	old M	?	Cystitis
Ralfe	1887	middle aged F	?	Cysto-pyelonephritis
Senator	1891	66 M	Yeast	Prostatic hypertrophy
Mueller	1892	60 M	<i>B. coli</i>	Prostatic hypertrophy; cystitis
Frisch	1896	64 M	<i>B. coli</i> ; yeast	Cystitis
Pere	1897	38 M	<i>B. coli</i>	Cystitis
Von Loghem	1905	65 M	<i>Proteus vulgaris</i>	Prostatic hypertrophy
Eve	1909	middle aged M	<i>B. coli</i>	Cystitis; ascending pyelonephritis
Sorensen	1910	60 M	<i>B. lactis aerogenes</i>	Prostatic hypertrophy; cystitis
		64 M	<i>B. coli</i>	Urethral stricture
Mullern-Aspegren	1921	73 M	Saccharomyces	Prostatic hypertrophy
Young	1922	76 F	<i>B. coli</i> ; yeast	Urethral stricture; cystitis; diverticulum of bladder
Walthard	1922	66 M	<i>B. coli</i> ; <i>B. lactis aerogenes</i>	Cystitis; hypertrophy of prostate
Riley and Bragdon	1937	52 M	Yeast	Urinary retention; cystitis
Arthur and Johnson	1948	56 F	<i>B. coli</i> ; fungus	Diverticulum of bladder; cystitis
Spring and Hymes	1952	31 F	<i>B. coli</i>	Cystitis

of hydrogen. Favre²⁴ in 1888 isolated a gas-forming bacterium at autopsy from a nondiabetic woman with cystitis and pyelonephritis. Gas was present in the bladder and ureters. Senator¹ first reported the association of pneumaturia with cystitis in a patient who was a diabetic. In his patient the urine was acid and had the odor of a fermented yeast mixture. The gas was largely carbon dioxide and the fermented urine contained considerable alcohol on distillation. Yeast cells were

TABLE 2 NON-GLYCOSURIC CASES REPORTED IN THE LITERATURE

Author	Year	Age and Sex	Organism	Pathology
Raciborski	1860	40 M	?	No: available
Keyes	1882	67 M	?	Prostatic hypertrophy
		68 M	?	Probable urinary fistula*
		77 M	?	Prostatic hypertrophy
Favre	1888	? F	<i>B. coli</i>	Cysto-pyelonephritis
Schow	1892	? ?	?	Cord bladder
Heyse	1893	35 F	<i>B. lactis aerogenes</i>	Cord bladder
Schnitzler	1894	46 F	<i>B. lactis aerogenes</i>	Cystitis
Kelly and MacCallum	1898	36 F	?	Renal calculi
Bacari	1899	38 F	<i>B. coli</i> ; <i>Staph aureus</i>	Cystitis
Wildbolz	1901	52 M	<i>B. lactis aerogenes</i>	Urethral stricture
Taussig	1907	34 F	<i>B. coli</i>	Cystitis
Adrian and Hamm†	1907	35 F	<i>B. coli</i>	Cystitis
		27 M	<i>B. coli</i>	Urethral stricture
Cealic and Ceocalteau	1914	32 F	<i>B. coli</i>	Urethral stricture cystitis
Thevenot and Leboeuf	1923	53 M	<i>B. Friedlander</i>	Cystitis
Mulsow and Gillies	1934	53 M	<i>Nocardia</i> ; strep. and staph.	Cystitis; prostatic hypertrophy

*This case is probably one of vesico-enteral fistula and perhaps should have been excluded. A postmortem was not done to confirm this.

†Reported four cases, but two were probably malignancies with vesico-enteral fistulae, and so are omitted.

Note: Some of the early reported cases of cystitis, bladder stone, and cysto-pyelonephritis were probably secondary to an obstructive uropathy.

found in the urine. Frisch's patient, a diabetic male 38 years old, had pneumaturia caused by a yeast cystitis.¹⁹

Kelly and MacCallum²⁵ in 1898 reviewed all the reported cases of pneumaturia and added one of their own. Nine out of the 16 had glycosuria. Obstruction to the lower urinary tract was present in the majority. Two had vesical paralysis following myelitis.

Eve³ stated that pneumaturia in a glucose-free urine may indicate a masked glycosuria. His patient had glycosuria before the onset of pneumaturia, but not during or after its disappearance. The cases of Sorensen¹ and Walthard⁵ confirmed this observation. Their patients were aglycosuric at the onset of pneumaturia and later became glycosuric. In Walthard's patient the gas produced seemed to be less after glycosuria was found.

Arthur and Johnson³⁰ collected all the cases of pneumaturia in the American literature. There were seven, including their own. Of these, three were in diabetics who had a complicating cystitis. We wish to report the eighth case, and the fourth in a diabetic with a complicating cystitis.

CASE REPORT

A 31-year-old woman with a history of diabetes mellitus of 12 years' duration came to the office of one of us (M.S.) on April 21, 1951. For three years she had been troubled with swelling of the feet of varying degree. She was taking 30 units of protamine zinc daily, but did not follow a diet. She had had poor vision for the past two to three years because of bilateral cataracts. Two brothers had diabetes.

The physical examination was normal except for slight hypertension, cataracts and very slight edema of the legs. The blood pressure was 160/90. The neurological examination was within normal limits. The fasting blood sugar was 205. The urine contained sugar graded 4 plus, and the albumin graded 1 plus.

The patient was advised to take ammonium chloride and a low salt diet. She was advised to take 50 units of NPH insulin daily. Her initial complaint disappeared in a few weeks. She was seen again on Sept. 22, 1951 for a check-up and minor complaints. No leg edema was present.

She was seen again two weeks later. Although improved, she complained of a bearing-down feeling at the end of urination and felt as though urination was incomplete. The urine contained sugar graded 4 plus, albumin graded 2 plus; many white blood cells and red blood cells were found on microscopic examination. Because of the suspicion of the presence of an early neurogenic bladder related to the diabetes, she was referred to one of us (J.H.) for urologic study.

In the course of the urologic examination on October 10, 1951, the patient volunteered the information that once daily during the past month, usually after work, she passed gas via the urethra at the end of urination. The gas was passed in an explosive manner and with a feeling of a bubbling sensation in the urethra.

A roentgenogram taken before cystoscopy showed the presence of gas in the bladder. The bladder contained 40 cc. of hazy residual urine. On cystoscopy, the bladder mucosa and trigone were seen to be reddened throughout. The bladder neck was red and edematous. The ureteral orifices were both normal. The patient felt the first urge to void when the bladder contained about 200 cc. of water and had marked urgency at about 300

cc. This was indicative of a mild hypertonic bladder associated with cystitis. No gas bubbles were seen being emitted from the ureteral openings. Catheters were passed to the pelvis of each kidney. Urine flow and split function indigo-carmin tests were normal. The urine was grossly clear from each kidney. A retrograde pyelogram was within normal limits.

The bladder urine showed the presence of albumin graded 1 plus, and 2 per cent sugar. The microscopic examination showed numerous white blood cells and about 15 red blood cells per high power field. The sugar had disappeared from the bladder and kidney urine specimens after they had stood overnight.

The culture of urine from the bladder revealed *Escherichia coli* in abundance (Dr. A. J. Weil). This strain showed gas formation from dextrose and lactose well above average. The culture of urine from the left kidney showed *Escherichia coli* with the same cultural behavior. From the urine of the right kidney a few colonies of *Bacillus alcaligenes faecalis* were cultured. This organism is not a producer of gas from carbohydrates; the significance of its presence is difficult to assess. Accidental contamination could not be excluded from the single examination. Antibiotics sensitivity tests of the organisms isolated from the urine of the bladder showed no sensitivity towards penicillin and bacitracin, a high degree of sensitivity towards aureomycin, chloromycetin, and terramycin, and sensitivity in high concentration only towards streptomycin and dihydrostreptomycin.

On October 14, 1951, in order to evaluate further her urologic status, the patient was seen again by one of us (J.H.). Cystoscopy after the patient had voided urine which appeared clear on inspection revealed the absence of residual urine. The bladder capacity and tone were normal. The mucosa was approximately normal in color; the trigone was moderately injected. The ureteral orifices were normal. Some edema of the bladder neck was noted. The urine was sugar free, and all the other tests were normal.

Starting October 18, 1951, the patient was given aureomycin, later followed by chloromycetin. The day after the institution of therapy, the pneumaturia disappeared completely. By the third day of treatment, urinary frequency had also disappeared. A relapse occurred two weeks after the therapy was discontinued. On November 24, 1951, she was given Gantrisin, followed by Mandelamine. The pneumaturia disappeared in 72 hours. She has remained symptom-free for two months. The examination of the urine for pus and blood cells has been negative.

COMMENT

A woman with diabetes and with a history of cystitis developed pneumaturia. An inflamed bladder on cystoscopy and gas in the bladder on roentgenogram confirmed the diagnosis. There was no history of prior instrumentation or abnormality of the urinary tract. Studies revealed the presence of *Escherichia coli* in the urine of the bladder and left kidney, with the complete disappearance of sugar in these specimens when they were allowed to stand overnight. The organism was found to be a fermenter of dextrose and lactose. It was sensitive to aureomycin, chloromycetin and terramycin.

In view of the presence of 40 cc. of residual urine on the first examination the possibility of an early diabetic cord bladder was considered as the origin of the cystitis. However, the presence of about normal bladder capacity and the first desire to void at about 200 cc. (250 cc. is normal), ruled out a cord bladder. We believe the small amount of residual urine found on the first examination was due to the presence of cystitis and edema of the bladder neck. (The gas may be produced in the bladder or kidneys. In our case, the gas was produced in the bladder, as shown on roentgenogram.) Residual urine in the bladder enabled the infecting organism to act on the glycosuric urine for long periods of time and thus produce gas; however, residual urine is not necessary for gas production. In our case, the pneumaturia continued even after the disappearance of the residual urine from the bladder.

The pneumaturia disappeared within 48 hours after institution of aureomycin therapy, but recurred within two weeks after the discontinuation of the therapy, probably because the aureomycin was not administered for a long enough period. Two days after the institution of Gantrisin, followed by Mandelamine, she became asymptomatic. At present, January 14, 1952, the patient is free of pneumaturia and associated urinary symptoms.

DISCUSSION

The presence of residual urine offers a favorable medium for the growth of bacterial and saccharomycelial organisms. The gas is usually produced in the bladder. In the kidney, unless obstruction with stasis is present, gas formation will not occur, as the urine passes down very rapidly from the kidneys to the bladder.

Analysis of the reported pneumaturia cases, and our own, shows that: 1, it occurs with about the same frequency in glycosuric and nonglycosuric patients; 2, since only 39 cases have been reported in the

literature, it probably occurs more frequently than has been reported; 3, the urine is usually acid, but neutral or ammoniacal when decomposition sets in; 4, *Escherichia coli*, *B. lactis aerogenes* and yeast are the most frequent organisms found on urine culture; and 5, the gas produced is usually odorless.

On analysis Mueller¹⁸ found the gas in diabetic urine to consist of carbon dioxide, 9.1 per cent; hydrogen, 57.3 per cent; nitrogen, 33.5 per cent; and methane, 0.79 per cent. As the urine stands, more carbon dioxide is formed, up to 19 per cent, and hydrogen decreases in amount as a result of the fermentation. Hydrogen forms the main bulk of the gas in diabetics. In nondiabetics the gas formed is mainly carbon dioxide.

The gas produced in glycosuric cases is due to fermentation with production of butyric and lactic acid. Distillation of the urine may yield considerable amounts of alcohol. As stated before, the presence of residual urine and glycosuria offers a favorable medium for the growth of organisms.

The gas produced in those cases without glycosuria is believed to be due either to proteolytic breakdown of a special kind of albumin which is present in the urine, or to an *Escherichia coli* strain which can form gas from the breakdown of normal albumin. The larger part of the gas formed in these instances consists of carbon dioxide. Another theory for the formation of gas in nonglycosuric urine was postulated by Mulsow and Gillies,³⁶ who stated that the acid urine from the kidneys acting upon bicarbonates in retained bladder urine may produce gas. Lastly, it may be caused by some non-reducing carbohydrate; by glucose in amounts too small to be detected by methods in use; by glucose that occurs only at intervals; or by glucose that may have been previously fermented out (Sorensen⁴). In view of the latter facts, every case of pneumaturia should be carefully studied for the presence of diabetes, even in the absence of sugar in the urine.

Pneumaturia does not result from ordinary ammoniacal fermentation of the urine or from putrefactive decomposition, because the hydrogen sulfide gas that is formed is soluble. Symptoms are not striking. There may be frequency because of the associated cystitis and the distention of the bladder with gas. The passage of gas causes no pain, but a tickling sensation, and usually occurs at the end of urination in an explosive manner. The urine is cloudy and may contain pus. Recovery is rapid with the use of the appropriate drugs or antibiotics, control of the diabetes, and the elimination of the residual urine, if present. Death is due to infection of other organs, e.g. pyelonephritis with uremia. The

incidence of pneumaturia must be much greater than the reported number of cases. This may be due to the fact that either it is not recognized or not reported. In our case, the patient mentioned incidentally that she had been passing gas for one month. The fact that we were cognizant of this condition enabled us to recognize and eliminate it.

SUMMARY

A case of pneumaturia in a diabetic woman, with cystitis, due to an infection with *Escherichia coli*, has been reported with a review of the literature.

Pneumaturia in itself is unimportant. It is significant as part of a symptom complex usually associated with urinary obstruction and infection, with or without diabetes. Antibiotics will not clear up the pneumaturia and the underlying infection unless the obstruction, if present, is eliminated and the diabetes controlled. A urine that is persistently negative for sugar in the presence of pneumaturia does not necessarily exclude diabetes, as the sugar may have been completely fermented by the infecting organism.

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