Review Article

Zinner syndrome: an updated pooled analysis based on 214 cases from 1999 to 2020: systematic review

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Abstract: Zinner syndrome (ZS) is a rare anomaly of the Wolffian duct. We searched for case reports and case series to identify the most effective methods for examination and treatment. The PubMed/Medline, Embase, and Scopus databases were searched. Our searches yielded 160 case reports/case series and included 214 patients. The mean age at diagnosis was 29.35 years. The R:L distribution was 1:1. The most common symptoms were: frequency (24.3%), dysuria (23.1%) and perineal pain (20.2%) in ZS patients with clinical symptoms. The diagnostic investigations used most frequently were magnetic resonance imaging (MRI) (67.8%) and ultrasonography (65.0%). Maximum flow rate on uroflowmetry, semen ejaculate volume, sperm count, and sperm motility were significantly lower in patients with ZS. Among 193 patients with treatment details, 65.8% underwent surgery; 9.8% underwent aspiration; 24.3% were followed with observation. Two patients died after surgery; all other patients remained asymptomatic after surgery. Fifty-two patients had complications and comorbidities. In patients with ZS, the most common symptoms are seminal vesicle cyst enlargement and compression of the bladder, ureter, and reproductive system. The diagnosis mostly depends on radiological examination. Surgery may be effective, but complications may occur.

Keywords: Zinner syndrome (ZS); symptom; diagnostic examinations; treatment; surgery; complication

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Introduction

Zinner syndrome (ZS) is a very rare anomaly, which is associated with an embryologic abnormality that develops in the distal portion of the mesonephric or Wolffian duct between the 4th and 13th gestational week. Incomplete migration of the ureteric bud from the proximal portion of the mesonephric duct may also result in a failure to meet with the metanephros. This failure interferes with the ureteric bud's key role in differentiation of the metanephric blastema, and ipsilateral renal agenesis will occur, in addition to atresia of the ejaculatory duct. This leads to unsatisfactory drainage, subsequent distension of the seminal vesicle, and creation of a cystic structure (1).

The condition was first described by Zinner in 1914 (*Figure 1*). The incidence of ZS is very low. In one study of children in Taipei (2), the incidence of seminal vesicles with ipsilateral renal dysplasia was 0.0046% (13/28,000). The clinical symptoms associated with this condition, which are nonspecific, include frequent dysuria,

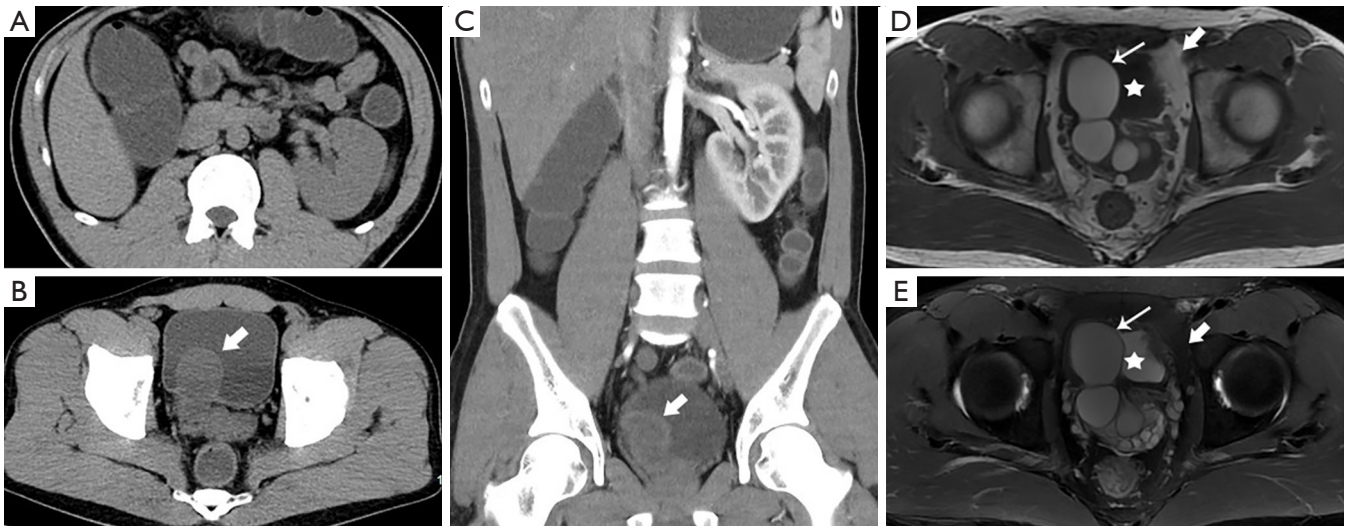

Figure 1 In a 30-year-old man with lower abdominal pain, CT presented the right renal agenesis (A) and posterior cystic lesions of the bladder (B,C, arrowheads). MRI showed the cystic lesions was attached to the right seminal vesicle and present as hyperintense in T1WI sequence and fat-suppression sequence (D,E) (short arrows) compared with bladder (stars) and fat (arrowheads). Operative and pathological findings as hemospermia in the seminal vesicle cyst. CT, computed tomography; MRI, magnetic resonance imaging.

epididymitis, perineal discomfort, and post-ejaculation pain. The severity of symptoms may correlate with the size of the seminal vesicle cyst (3). van den Ouden *et al.* (1) conducted a pooled analysis of patients with ZS in 1998. The study provided important reference information for clinicians and researchers; however, as times change and more cases are reported, an updated review of ZS cases may be necessary. We therefore performed a comprehensive review, which included case reports and case series published during the period from 1999 to 2020. We present our article in accordance with the PRISMA reporting checklist (available at http://dx.doi.org/10.21037/apm-20-1997).

Methods

A literature search was performed using the PubMed/Medline, Embase, and Scopus databases and the language was no restrictions. Two physicians with literature retrieval experience performed the search using the following keywords: zinner syndrome; zinners syndrome; seminal vesicle cyst; ipsilateral renal agenesis; ipsilateral renal hypoplasia. Studies published during the period from January 1st, 1999 to May 1st, 2020 were included in the study. The data search was limited to case reports and case series. In order to identify other potentially eligible publications, the references from the articles identified initially were manually reviewed. The diagnosis of ZS was considered in cases with of seminal vesicle cyst formation combined with ipsilateral renal agenesis.

The case reports were analyzed with regard to the data provided on patient age, clinical symptoms, site of pathogenesis, urogenital function, results on diagnostic examinations, treatment options, duration of follow-up, prognosis, complications, and comorbidities. The research is being reported in line with the attached PRISMA reporting checklist.

Statistical analysis

Descriptive statistics were used to summarize the variables in the case reports and case series.

Results

In total, 160 case reports and case series (Table S1) were obtained from the PubMed/Medline, Embase, and Scopus databases (*Figure 2*). All these cases met the proposed diagnostic criteria for ZS, and 214 patients were ultimately included.

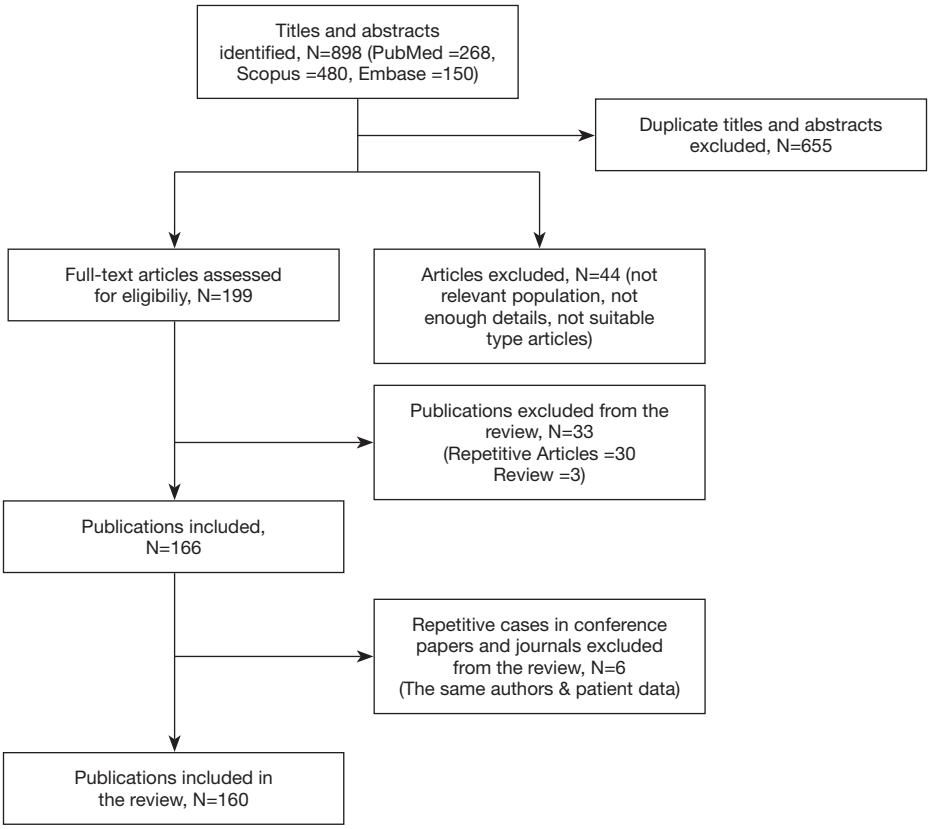

Figure 2 Flow-chart depicting the screening process.

Patient characteristics at baseline

Twenty-one patients were removed due to a lack of data (4,5). The age of patients included in the study ranged from newborn (6) to 76 years (7) (mean age: 29.35 years) (*Table 1*). Of 214 patients, 173 (80.8%) had clinical symptoms, while the remaining cases were detected incidentally without symptoms (163%, 35/214), or had symptoms that were not described in detail (3.3%, 7/214). The symptoms described in the included studies were diverse, and most patients presented with multiple symptoms. Most patients presented with urinary symptoms, including: urinary frequency (24.2%, 42/173); dysuria (26.0%, 45/173); urinary urgency (13.9%, 24/173); micturition pain (8.2%, 14/173), hematuria (7.5%, 13/173); nocturia (1.7%, 3/173); urinary incontinence (1.2%, 2/173). The other common symptoms reported included: local pain, including perineal pain (20.2%, 35/173); abdominal pain (14.5%, 25/173); pelvic pain (6.9%, 12/173); suprapubic pain (4.0%, 7/173); backache (3.5%, 6/173); inguinodynia (2.9%, 5/173); testicular pain (1.7%, 3/173); scrotal pain (1.2%, 2/173); coxalgia (0.6%, 1/173). Another category of symptoms in this patient population comprised abnormalities of reproductive function, including: infertility (13.9%, 24/173); ejaculation-related pain (11.0%, 19/173); hematospermia (8.7%, 15/173); testicular swelling (3.5%, 6/173); reduced or negligible semen volume (2.9%, 5/173); weak ejaculation (2.9%, 5/173); sexual discomfort (2.3%, 4/173); erectile dysfunction (1.2%, 2/173). Other symptoms reported included: constipation (4.0%, 7/173), fever (3.5%, 6/173), pain on defecation (1.2%, 2/173), loss of weight (1.2%, 2/173), nausea and vomiting (1.2%, 2/173), and so on. All reported symptoms are summarized in *Table 2* and *Figure 3*. In cases with seminal vesicle cyst enlargement and compression of the bladder, the most significant symptoms may be dysuria, perineal pain, urinary frequency, and urinary urgency. In 110 patients (51.4%), the seminal vesicle cyst and renal agenesis were present on the patient's right side. In 104 patients (48.6%), the seminal vesicle cyst and renal agenesis were present on the patient's left side. Thus, the patient's right side was more commonly affected, but the R:L ratio was approximately 1:1.

Diagnostic examinations

Table 3 summarizes the results of diagnostic examination for the 214 patients included in the study. Most patients underwent more than two diagnostic examinations. The techniques used most commonly for diagnosis were: magnetic resonance imaging (MRI) (145/214, 67.8%), ultrasonography (139/214, 65%), computed tomography (CT) (130/214, 60.7%), urography (41/214, 19.2%), and digital rectal examination (DRE) (40/214, 18.7%).

Although radiology was most commonly used for the diagnostic examination, cystoscopy (30/214, 14.0%) and transrectal ultrasonography (TRUS) (29/214, 13.6%) were used to obtain additional information. Diagnostic methods such as retrograde pyelography (7/214, 3.3%) (8-14), radioactive nephrography (5/214, 2.3%) (9,15-18), positron emission tomography-computed tomography (PET-CT) (2/214, 0.9%) (19,20), and digital subtraction angiography (DSA) (2/214, 0.9%) (21,22) were effective in special situations.

Table 1 Distribution of age (in decades) at the time of presentation in 193 ZS patients

Decade (year)	N	%
0–10	23	11.9
11–20	32	16.6
21–30	64	33.1
31–40	28	14.5
41–50	23	11.9
51–60	10	5.2
61–70	12	6.2
71–80	1	0.5

ZS, Zinner syndrome.

Effects on urinary and reproductive function

When a seminal vesicle cyst compresses vital structures, maximum flow rate, as measured by uroflowmetry, may be attenuated. Uroflowmetry was performed in 10 cases. The maximum flow rate was 3.1 (18) to 16 (23) m/s, with median and standard deviation of 10.4 and 4.2 m/s, respectively. The normal reference range for maximum flow rate is ≥15 m/s.

Ejaculate volume was measured in 12 patients. Values

Table 2 Summary of symptoms in 173 ZS patients

Urinary symptoms		Local pain		Abnormal of reproductive function		Other symptoms	
Symptoms	Number	Symptoms	Number	Symptoms	Number	Symptoms	Number
Frequency	42	Perineal pain	35	Infertility	24	Constipation	7
Dysuria	45	Abdominal pain	25	Ejaculation pain	19	Fever	6
Urgent	24	Pelvic pain	12	Hematospermia	15	Defecation pain	2
Micturition pain	14	Suprapubic pain	7	Swelling of testicles	6	Loss of weight	2
Hematuresis	13	Backache	6	Ejaculation weakness	5	Nausea and vomiting	2
Nocturia	3	Inguinodynia	5	Less or no semen	5	Diarrhea	1
Urinary Incontinence	2	Testicular pain	3	Sexual discomfort	4	Gastrointestinal uncomfortable	1
		Scrotal pain	2	Erectile dysfunction	2	Fatigued	1
		Coxalgia	1			Multiple	1
						Fremitus	1
						Abdominal distension	1
						Night sweats	1
						Malaise	1

ZS, Zinner syndrome.

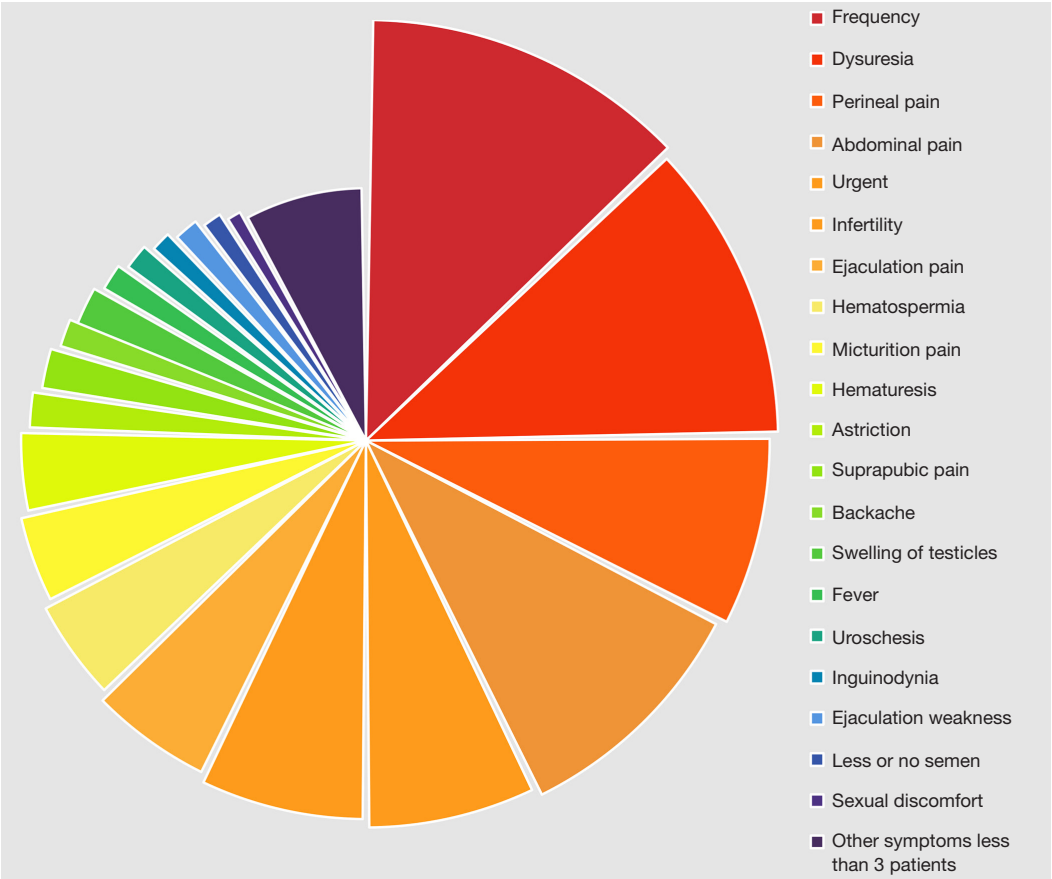

Figure 3 The frequency of various symptoms in 173 ZS patients. ZS, Zinner syndrome.

ranged from 0.1 (24) to 2.2 (25) mL, with median and standard deviation of 1 and 0.6, respectively. The normal reference range for ejaculate volume is 2–6 mL. Details relating to sperm count were available for 7 patients. Among the patients included in this study, sperm count ranged from 160,000 (26) to 11,000,000 (27), with median and standard deviation of 1,200,000 and 5,200,000, respectively. The normal reference range for sperm count is >20,000,000. Data on sperm motility were available for 7 patients. The percentage of non-motile sperm in a given patient ranged from 59.5% (28) to 100% (24), with median and standard deviation of 90% and 18.35%, respectively. In an additional 12 cases, patients had were reported to have azoospermatism and oligospermia, but details were not provided.

Treatment and management

Some of the cases included in this study did not describe treatment. Details related to treatment and management were available for 193 patients (*Table 4*). Surgery, which was the most common approach to treatment, was performed in 127 cases (65.8%, 127/193), including 39 patients who underwent open surgery, 23 patients who underwent laparoscopic surgery, 23 patients who underwent robot-assisted laparoscopic vesiculectomy, five patients who underwent seminal vesiculoscopy, four patients who underwent transurethral resection of the ejaculatory duct, one patient who underwent ultrasound-guided transrectal biopsy, one patient who underwent interventional surgery, one patient who underwent cystoscopy and aspiration of the cyst, one patient who underwent transurethral unroofing of the cyst, and 29 patients who underwent surgery, with no details provided. In one case, death was caused by the metastasis of a papillary adenocarcinoma in the seminal vesicle after radical cystoprostatectomy (24). In another case, death was caused by multiple organ failure related to renal-type clear cell carcinoma in the prostate after surgery (29).

Table 3 Summary of diagnostic examination results for 214 ZS patients

Diagnostic examinations	N	The percentage of choice in 214 patients (%)
MRI	145	67.8
Trans-abdominal ultrasound	139	65.0
CT	130	60.7
Urography	41	19.2
DRE	40	18.7
cystoscopy	30	14.0
TRUS	29	13.6
Retrograde pyelography	7	3.3
Radioactive nephrogram	3	1.4
PET-CT	2	0.9
99mTc-DMSA	2	0.9
DSA	2	0.9

ZS, Zinner syndrome; MRI, magnetic resonance imaging; CT, computed tomography; DRE, digital rectal examination; TRUS, transrectal ultrasonography; PET-CT, positron emission tomography-computed tomography; DSA, digital subtraction angiography.

The other patients who underwent surgery did not experience symptom recurrence or other complications.

With regard to invasive procedures, 19 patients (19/193, 9.8%) underwent aspiration and drainage; 18 patients underwent transrectal aspiration of the cyst, and one patient underwent percutaneous drainage under ultrasound guidance. Nine patients (9/19, 47.4%) had cyst recurrence requiring surgical treatment, and one patient died of sepsis caused by an infection of the seminal vesicle cyst (30).

Forty-seven patients elected to receive conservative treatment and follow-up because of a lack of symptoms, the presence of only minor symptoms, or personal reasons (24.2%, 47/193). The range of follow-up time ranged from 3 months to 15 years. During follow-up, an increase in seminal vesicle cyst size necessitated surgery in 2 cases (31,32). Vasectomy was performed in one infant for recurrent epididymitis and orchitis. Nephrectomy was performed in three infants for unilateral multicystic dysplastic kidney. Kidney transplantation was performed in three infants for the treatment of bilateral dysplastic kidney (6).

Complications and comorbidities

In most cases, ZS is associated with a good prognosis. However, among the cases included in this study, 52 patients had complications or comorbidities, and three died (*Table 5*, *Figure 4*). According to the definitions of complications and comorbidities, as well as reviews of the literature and discussions with urologists and radiologists, we treated urogenital infection and inflammation as complications and treated tumors, developmental deformities, and other diseases as comorbidities. The urogenital infection and inflammation in collection cases including urinary infection (13 patients), epididymitis (6 patients) (6,33-37), prostatitis (2 patients) (38,39), orchitis (2 patients) (6,13), bladder infection (1 patient) (6), and sepsis caused by an infection of the seminal vesicle cysts, resulting in death (1 patient) (30). The most common comorbidities were the development of tumors, including adenoma or adenocarcinoma (40-45) arising from the seminal vesicle cyst (6 patients), paraganglioma affecting the bladder in 1 patient and affecting paranephrotic tissue in another patient (46,47), squamous cell carcinoma arising from the seminal vesicle (1 patient) (48), non-hodgkin lymphoma (1 patient) (19), and renal-type clear cell carcinoma affecting the prostate (1 patient) (43). Two patients with adenocarcinoma associated with a seminal vesicle cyst died because of metastasis and multiple organ failure. ZS may also be combined with other developmental deformities, including cryptorchidism (2 patients) (49,50), anorectal malformation (2 patients), contralateral dysplastic kidney (2 patients), ventricle septum defect (2 patients) (6), situs inversus viscerum (1 patient) (51), mitral insufficiency (1 patient) (52), and multiple severe developmental deformities (1 patient) (53). Comorbidities caused by unrelated illness or syndromes were relatively rare. In this study, 3 patients had complications related to Kallmann syndrome (21,54-56), 1 patient had complications related to thalassemia, and 1 patient had complications related to Fabry disease (57).

Discussion

ZS is a rare condition. The constellation of associated congenital findings includes ipsilateral renal hypoplasia, cystic seminal vesicles, and ejaculator duct obstruction. It occurs with anomalous growth of the mesonephric or

Table 4 Summary of management in 193 ZS cases

Management measures	N	%	Prognosis
Open operation	39	20.2	1 patient dead
Surgical operation no details	29	15.0	1 patient dead
Laparoscopic surgery	23	11.9	Favorable
Robotic-assisted laparoscopic vesiculectomy	23	11.9	Favorable
Seminal vesiculoscopy	5	2.6	Favorable
Transuretral resection of the ejaculatory duct	4	2.1	Favorable
Ultrasound-guided transrectal biopsy	1	0.5	Favorable
Interventional operation	1	0.5	Favorable
Cystoscope and aspiration of the cyst	1	0.5	Favorable
Transurethral unroofing of the cyst	1	0.5	Favorable
Aspiration and drainage	19	9.8	9 patients recurrence & 1 patient dead
Follow up	47	24.3	9 patients (included 7 infants) need surgery
Total	193	100.0	

ZS, Zinner syndrome.

Wolffian duct between 4 and 13 weeks of gestation and negatively impacts embryological development of the kidney and ejaculatory duct.

A pooled analysis was published by van den Ouden *et al.* in 1998. We sought to provide an updated review to help clinicians. Most patients with ZS present during the second or third decade of life. Compared with the study published by van den Ouden *et al.*, our study had a greater sample size. We found that there was no obvious difference in the incidence of left-sided *vs.* right-sided disease. This finding differs from those reported by van den Ouden *et al.* The most common symptoms among our patient population were dysuria, perineal pain, urinary frequency, and urinary urgency. These symptoms may be associated with an increase in cyst size. On the other hand, some patients had impaired reproductive function, including 12 patients presented with low-volume ejaculation, seven patients had low sperm motility, and 19 patients presented with azoospermatism and oligospermia. Decreases in sperm count may be related to obliteration of the ejaculatory duct, the hemorrhage of seminal vesicle cysts, or secondary inflammation of the genitourinary system.

We also found that the choice of diagnostic examination had important effects on the selection of radiological techniques. In 1998, DRE and intravenous urography were commonly used for the diagnosis of ZS. In contrast, the studies included in this review used MRI, trans-abdominal ultrasound, and CT because of these techniques' noninvasive nature and ease of use. MRI was recommended by radiologists to differentiate between lesions and surrounding normal tissue as well as the effects of non-ionizing radiation. Recent developments in the field of MRI have led to improved field intensity and new sequences, which provide more information about the condition of underdeveloped kidneys and ureters, cyst contents, and surrounding tissue or organs affected by complications. This information is helpful for guiding the clinical decision. In this review, seminal vesicle cyst and ipsilateral renal agenesis were first detected by trans-abdominal ultrasound because of its convenience for screening. CT is an important supplement for patients with contraindications for MRI. The information obtained via CT and urography can supplement other information such as the extent of calcification and urinary function.

In the studies included in this review, surgery (open or laparoscopic) was the most effective treatment and postoperative recurrence was rare. As new methods of surgery that were not mentioned in the study by van den Ouden *et al.*, endoscopy and robot-assisted technology have become increasingly popular among physicians performing seminal vesiculectomy since 2002 (58) and 2007 (59), respectively. Compared with open seminal vesiculectomy,

Table 5 Complications and comorbidities in 52 ZS patients

Complications and comorbidities	N	The percentage in 214 patients (%)
Tumors	11	
Adenocarcinoma (seminal vesicle)	5	2.34
Adenoma	1	0.47
Paraganglioma (bladder and aranephros)	2	0.93
Squamous cell carcinoma (seminal vesicle)	1	0.47
Non-Hodgkin lymphoma (lymphoid tissue)	1	0.47
Renal-type clear cell carcinoma (prostate)	1	0.47
Developmental deformities	10	
Situs inversus viscerum	1	0.47
Mitral insufficiency	1	0.47
Cryptorchidism	2	0.93
Anorectal malformation	2	0.93
Contralateral dysplastic kidney	2	0.93
Ventricle septum defect	2	0.93
Multiple developmental deformity	1	0.47
Infections or inflammations	25	
Urinary infection	13	6.07
Bladder infection	1	0.47
Epididymitis	6	2.80
Prostatitis	2	0.93
Orchitis	2	0.93
Sepsis	1	0.47
Other illness or syndromes	5	
Kallmann syndrome	3	1.40
Thalassemia	1	0.47
Fabry disease	1	0.47
Total number of patients	52	23.83

ZS, Zinner syndrome.

laparoscopic and robot-assisted seminal vesiculectomy are safer, less invasive, accompanied by less bleeding, and associated with quicker recover. Among the 46 patients included in this review who underwent laparoscopic (23 patients) or robot-assisted seminal vesiculectomy (23 patients), prognosis was favorable, with no postoperative complications reported. However, our sample size was insufficient to demonstrate the safety of surgical treatment for ZS. A large-scale of meta-analysis (60) that included more than 200,000 patients indicated that open retropubic radical prostatectomy (ORP), laparoscopic radical prostatectomy (LRP), and robot-assisted laparoscopic radical prostatectomy (RALP) were associated with high risk for surgical complications leading to erectile dysfunction (ORP: 42.7–61.8%; LRP: 45–71.1%; RALP: 23.4–68.2%) and urinary incontinence (ORP: 15.4–28%; LRP: 7.1–42.8%; RALP: 7.7–20.7%). Although the target organs are different, the surgical methods, surgical approaches, and surrounding tissues are similar between prostatectomy and seminal vesicle resection, seminal vesicle resection still carries risk for surgical complications. Postoperative recurrence was not found in the van den Ouden *et al.* (1) report or our population, but that does not mean that it is impossible. Additional cases and longer follow-up time are necessary.

Another option in such cases is aspiration and drainage. van den Ouden *et al.* reported that the rate of success after aspiration was 30%, and 47.4% of patients included in the present review who underwent aspiration had symptom recurrence. As a variety of invasive treatment, aspiration was unable to decrease the risk for seminal vesicle cyst recurrence, so it is not valuable as a management strategy. Asymptomatic patients should be followed with observation, but when symptoms appear, clinical intervention is needed.

The complications and comorbidities experienced by ZS patients deserve attention but have been neglected in the past. In this review, three patients died because of complications or comorbidities. Secondary inflammation and ZS-related are the frequent complications in this patient population. These complications present with symptoms, so they should not easily be overlooked. However, tumor as comorbidity may not present with any specific symptoms in the early stage. In our review, tumors occurred in 10 patients (4.4%), and two patients died because of multiple metastases or organ failure. Patients with ZS and tumors arising from the urogenital system [including adenocarcinoma of the seminal vesicle (5, 2.34%), clear cell renal carcinoma of the prostate (1, 0.47%), paraganglioma (2, 0.93%) and squamous cell carcinoma of the seminal vesicle (1, 0.47%)] are rare conditions in ordinary people. According to the relatively high incidence of disease in this ZS population, although the exact reason remains unknown, ZS may be a positive stimulating factor for urogenital system tumors. Other developmental abnormalities or diseases can occur, but they are relatively rare. Remarkably,

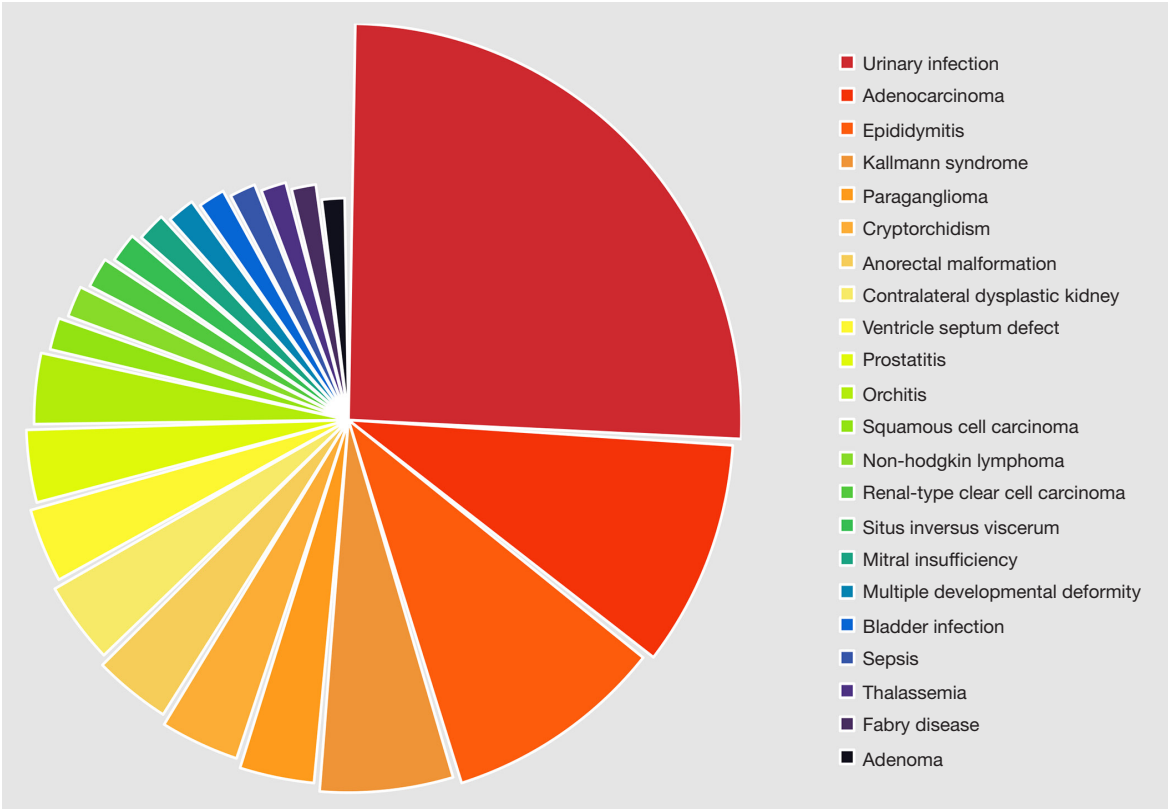

Figure 4 The frequency of complications and comorbidities in 52 ZS patients. ZS, Zinner syndrome.

three ZS patients included in this review had Kallmann syndrome, which was more common than cryptorchidism and contralateral dysplastic kidney. We consider that ZS and Kallmann syndrome may have some kind of relationship, but the cases and data were not sufficient for investigation.

This review has several limitations. First, it was not possible to classify patients by ethnicity because the information was lacking from many studies. Only 13 patients could be classified according to ethnicity: 7 Caucasian males; 2 Saudi Arabian males; 1 Indian male; 1 Hispanic male; 1 Moroccan male; 1 Italian male. Second, it was not possible to classify seminal vesicle lesions based on wall changes because the information was lacking from many studies. Third, there may have been some publication bias, resulting in overestimations of the proportions of symptoms, complications, and comorbidities. Fourth, a majority of cases lack relevant information such as uroflowmetry, semen ejaculate volume, sperm count, and sperm motility, so it difficult to provide convincing data on the effect of ZS on male reproductive function. Finally, some of the studies and conference papers identified by our initial literature search provided only abstracts. We had to exclude these publications from our review. In conclusion, the clinical symptoms associated with ZS syndrome are varied, but the most common symptoms are enlargement of seminal vesicle cysts and compression of the bladder, ureters, and reproductive system, as well as associated secondary lesions. The diagnosis mostly depends on the results of radiological examinations. Complications and comorbidities should be identified, with treatment tailored accordingly.

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Footnote

Reporting Checklist: The authors have completed the PRISMA reporting checklist. Available at http://dx.doi.org/10.21037/apm-20-1997

Conflicts of Interest: All authors have completed the ICMJE

uniform disclosure form (available at http://dx.doi.org/10.21037/apm-20-1997). The authors have no conflicts of interest to declare.

Ethical Statement: The authors are accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved.

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Table S1 Distribution, clinical symptoms and ages features of Zinner syndrome (ZS) from 160 case report/case series

No.	Author	Number & distribution (R = right, L= left)	Age (y = year, m = month)	Symptoms and complains
1	Landry JL (61)	1R	24 y	Perineal pain
2	Steffens J (62)	1R	10 m	Asymptom
3	Livingston L (63)	3L & 2R	17–60 y	Dysuresia [4], perineal pain, urgent, frequency [2], infertility [2], nocturia, hematuresis, hematospermia, abdomen pain & ejaculation pain
4	Gorrea M (64)	1R	37 y	Hematuresis
5	Eberli D (65)	1R	9 y	Swelling of Testicles
6	Burns JA (8)	1R	12 y	Swelling of Testicles
7	Buoggi G (58)	1L	23 y	Inguinodynia & sexual discomfort
8	Manousakas T (66)	1L	30 y	Hematuresis, frequency & urgent
9	Radhia S (67)	1R	34 y	Dysuresia, frequency, urgent & pelvic pain
10	Takada S (68)	1L	39 y	Asymptom
11	Calahorra Fernández FJ (26)	1R	41 y	Hematospermia & infertility
12	Giglio M (33)	1L	45 y	Epididymitis
13	Roberts SG (69)	1R	49 y	Asymptom
14	Kluckert JT (40)	1L	67 y	Dysuresia
15	Hoschke B (70)	1L	14 y	Asymptom
16	Protogerou V (71)	1L	24 y	Frequency, urgent, backache,inguinodynia
17	Segawa N (72)	1R	29 y	Uroschesis & perineal pain
18	Akao J (73)	1R	29 y	Dysuresia
19	Narlawar RS (74)	1R	30 y	Dysuresia, frequency & ejaculation pain
20	Murphy JO (75)	1L & 1R	15y & 22y	Dysuresia [2] & suprapubic pain [1]
21	Basillote JB (76)	1R	7y	Dysuresia & suprapubic pain
22	Kilinç F (77)	1L	17 y	Dysuresia & perineal pain
23	Liatsikos EN (34)	1L	26 y	Dysuresia, frequency & urgent
24	Kawahara T (24)	1R	30 y	Infertility
25	Pascual Samaniego M (78)	1R	34 y	Inguinodynia
26	Madrid Garcia FJ (79)	1L	42 y	Dysuresia & frequency
27	Berg F (35)	1R	38 y	Perineal pain & ejaculation pain
28	Chuang KL (15)	1R	53 y	Abdominal pain
29	Kosan M (80)	1L	20 y	Hematuresis
30	Gözen AS (81)	1R	22 y	Dysuresia, frequency & uracratia
31	Romero Selas E (82)	1R	26 y	Asymptom
32	Andrade-Rocha FT (25)	1R	39 y	Infertility
33	Pal D (83)	1L	51 y	Dysuresia
34	Cihan A (84)	1L	61 y	Asymptom
35	Chen HW (85)	4L & 2R	10–15 y	Asymptom [4], dysuresia, frequency & urgent [1]
36	Savica V (21)	1R	13 y	Coxalgia
37	Moore CD (59)	1R	16 y	Nausea, vomiting, abdominal pain & swelling of testicles
38	Kondo N (41)	1R	30 y	Hematospermia, perineal pain, micturition pain
39	Suárez Aliago B (86)	1L	33 y	Abdominal pain & fever
40	Lee BH (42)	1L	41 y	Hematuresis
41	Han P (87)	1L	41 y	Perineal pain & hematospermia
42	Carbone A (88)	1 & 1R	24 y & 43 y	Abdominal pain & infertility [1], no semen [1]
43	Pace G (4)	7R	32 y (mean)	Infertility [7], ejaculation weakness [5], dysuresia [4], ejaculation pain [4] & perineal pain [4]
44	Lilje C (53)	1R	5 m	Multiple developmental malformation
45	Labanaris AP (89)	1R	27 y	Sexual discomfort, frequency & urgent
46	Casey RG (90)	1R	32 y	Swelling of testicles & testicular pain
47	Štimac G (91)	1R	36 y	Inguinodynia, frequency & urgent
48	Ohgaki K (92)	1L	38 y	Perineal pain & micturition pain
49	Selli C (93)	1R	39 y	Perineal pain &dysuresia
50	Kowalczyk K (52)	1R	3 y	Swelling of testicles
51	Sotelo R (94)	1R	19 y	Micturition pain, frequency & urgent
52	Pereira BJ (95)	1R	22 y	Dysuresia, perineal pain & ejaculation pain
53	Ulu EMK (16)	1R	25 y	Asymptom
54	Chung SD (30)	1R	56 y	Fever, dysuresia& uroschesis
55	Seo IY (96)	4L	22 y, 26 y, 67 y & 68 y	Perineal pain [2] & hematospermia [2]
56	Schulkeh N (6)	5L & 4R	Newborn to 50 m	Asymptom [8], epididymitis and orchitis [1]
57	Ghonge NP (97)	1R	19 y	Dysuresia, frequency, perineal pain, scrotal pain & ejaculation pain
58	Sukumar S (9)	1R	20 y	Abdominal pain
59	Lim Christopher SH (98)	1R	23 y	Ejaculation pain
60	Viktorsdóttir MB (99)	1R	25 y	Perineal pain
61	Allaparthi S (100)	1R	34 y	No mention
62	Dombován P (101)	1L	34 y	Abdominal pain
63	Kao CC (28)	1L	43 y	Perineal pain & infertility
64	Ahallal Y (102)	1L	16 y	Micturition pain, frequency & urgent
65	Hampton LJ (103)	1L	35 y	Micturition pain, less or no semen & epididymitis
66	Kovac JR (104)	1L	36 y	Abdominal pain, astriction, nausea, vomiting & fever
67	Jang KD (105)	1L	49 y	Frequency, nocturia, & dysuresia
68	Kuo J (19)	1L	52 y	Fever, loss of weight & night sweats
69	Kalyanraman B (106)	1L	20 y	Perineal pain, dysuresia & defecation pain
70	Devaraju S (107)	1R	28 y	Abdominal pain, dysuresia, frequency & suprapubic pain
71	Basal S (108)	1L	28 y	Perineal pain & dysuresia
72	El-Assmy A (109)	1R	34 y	Frequency & nocturia
73	Haghighi R (110)	1L	36 y	Perineal pain & hematospermia
74	Fujita M (7)	1R	76 y	Dysuresia
75	Heller MT (36)	2R	46 y, 27 y	Perineal pain [2]
76	Coşkun B (111)	1L	20 y	Uracratia, pelvic pain, sexual discomfort & hematospermia
77	Kardoust Parizi M (112)	1L	27 y	Hematospermia, ejaculation pain, infertility, micturition pain, frequency & urgent
78	Sridhar AN (54)	1R	36 y	Kallmann syndrome, hematuresis & hematospermia
79	Nayak B (10)	1R	44 y	Dysuresia, hematuresis & frequency
80	Altobelli E (113)	1L	66 y	Fever & fremitus
81	Silveri M (114)	1L	2 y	Abdominal pain & astriction
82	Batur A (115)	1L	22 y	Pelvic pain & hematospermia
83	Royston E (22)	1L	23 y	Ejaculation pain
84	Palmer WC (116)	1L	26 y	Fever and pelvic pain
85	Satyanarayana R (43)	1R	29 y	No mention
86	Kim YH (48)	1R	29 y	Less or no semen & hematuresis
87	Lee KH (117)	1L	46 y	Abdominal pain, hematuresis, dysuresia, micturition pain, frequency & urgent
88	Mcadams S (38)	1R	54 y	Dysuresia, frequency & pelvic pain
89	Shepherd G (118)	2L	18 m & 2 y	Asymptom [2]
90	Kanavaki A (119)	1R	4 y	Asymptom
91	Burak Özkan M (120)	1L	15 y	Pelvic pain
92	Haddock P (11)	1R	18 y	Ejaculation pain
93	Valsangkar RS (46)	1R	21 y	Hematuresis
94	Donato P (121)	1L	22 y	Abdominal pain & diarrhea
95	Sundar R (12)	1L	25 y	Frequency, pelvic pain & ejaculation pain
96	Alp B (122)	1L	28 y	Frequency & pelvic pain
97	Harbo FS (20)	1L	41 y	Swelling of testicles
98	Kim Y (123)	1R	41 y	Hematuresis
99	Pavan N (124)	1R	42 y	Asymptom
100	Juho YC (125)	1R	43 y	Gastrointestinal uncomfortable, less or no semen & frequency
101	García Asensio D (126)	1L	10 m	Asymptom
102	Domínguez A (55)	1R	20 y	Kallmann symdrome, testicular pain & erectile dysfunction
103	Leen A (56)	1L	22 y	Kallmann syndrome & testicular hypoplasia
104	Jaiswal A (17)	1R	25 y	Micturition pain, frequency & urgent
105	Mehra S (27)	1R	25 y	Dysuresia, frequency & infertility
106	Aghawayi I (127)	1L	28 y	Abdominal pain
107	Hajji F (128)	1L	29 y	Dysuresia, urgent, abdominal pain, sexual discomfort & hematospermia
108	Dogan F (129)	1R	38 y	Hematospermia, dysuresia, astriction & pelvic pain
109	Shah S (130)	1R	39 y	Uroschesis & suprapubic pain
110	Baklouf F (131)	1R	43 y	Infertility
111	Gutierrez García MA (132)	1L	46 y	Pelvic pain & ejaculation pain
112	Deptala A (44)	1L	58 y	Frequency, backache, loss of weight & astriction
113	Maré A (133)	1R	62 y	No mention
114	Sato Y (29)	1L	64 y	Hematuresis
115	Yu YD (49)	1R	10 m	Asymptom & undescended testis
116	Slaoui A (134)	1L & 1R	35 y & 39 y	Hematuresis [1], backache [1] & no mention [1]
117	Canales-Casco N (135)	1L	17 y	Hematospermia
118	Prem G (136)	1L	19 y	Dysuresia, frequency, urgent & ejaculation pain
119	Chiapparrone G (137)	1L	20 y	Perineal pain & infertility
120	Cao JD (138)	1L	23 y	Perineal pain
121	Ahmed A (139)	1L	30 y	Backache & abdominal pain
122	Campora M (140)	1R	37 y	Asymptom
123	Khanduri S (141)	1R	40 y	Abdominal pain, perineal pain, ejaculation pain & infertility
124	Fiaschetti V (51)	1L	56 y	Perineal pain, dysuresia, frequency & less or no semen
125	Naval-Baudin P (50)	1L & 1R	26 y & 29 y	Testicular pain [1] & infertility [1]
126	Roth C (142)	1L	15 y	Perineal pain & frequency
127	Miliman AL (143)	1R	21 y	Frequency, urgent & perineal pain
128	Maehana T (18)	1R	21 y	Dysuresia
129	Razdan S (144)	1L	21 y	Abdominal pain, pelvic pain & astriction
130	Florim S (145)	1R	21 y	Scrotal pain
131	Kord E (146)	1L	23 y	Micturition pain, frequency, urgent & perineal pain
132	Kiremit MC (23)	1L	23 y	Micturition pain, frequency, urgent & perineal pain
133	Siguenza Gonzalez R (39)	1R	23 y	No mention
134	Farooqui A (147)	1R	28 y	Malaise, fatigue, & myalgia
135	Brown M (148)	1L	31 y	Micturition pain, frequency & urgent
136	El Mortaji H (149)	1R	45 y	Backache
137	Ali MS (47)	1L	67 y	Micturition pain, frequency, urgent & ejaculation pain
138	Guan J (5)	6L & 8R	14–43 y	Hematospermia [1], infertility [2], abdominal pain [3], ventosity [1], frequency & dysuresia [7]
139	Briosa F (150)	1R	14 y	Asymptom
140	Sousa Célia (57)	1R	17 y	No mention
141	Symeonidis EN (151)	1L	19 y	Asymptom
142	Kori R (152)	1R	19 y	Dysuresia
143	AlArifi M (153)	1L	20 y	Inguinodynia, testicular pain & ejaculation pain
144	Aslan Serdar (154)	1L	27 y	Pelvic pain
145	Cito G (155)	1L	27 y	Perineal pain & dysuresia
146	Bhat A (45)	1R	28 y	Perineal pain & abdominal pain
147	Bhukte S (156)	1L	28 y	Abdominal pain
148	Patil M (157)	1R	32 y	Infertility
149	Sahare P (158)	1R	45 y	Micturition pain, frequency, urgent & dysuresia
150	Alhajeri F (159)	1L	46 y	Testicular pain, suprapubic pain & uroschesis
151	Corongiu E (160)	1L	51 y	Perineal pain, astriction, infertility
152	Chibelean BC (161)	1L	61 y	Abdominal pain & astriction
153	Cascini V (13)	1L & 1R	1 m & 1 m	Asymptom
154	Herrero Blanco E (37)	1L & 1R	22 y, 29 y	Asymptom [2]
155	Bryson CF (162)	1R	24 y	Abdominal pain, suprapubic pain, testicular pain and frequency
156	Hergan B (163)	1L	22 y	Asymptom
157	Yadav SP (164)	1L	29 y	Suprapubic pain, uroschesis,
158	Tan Z (165)	2L & 4R	15–64 y	Micturition pain [1], Frequency [2], Urgent [2] Backache [1], Asymptom [2], hematochezia [1]
159	te Dorsthorst MJ (31)	2L& 1R	56 y, 61 y, 67 y	Asymptom [1], erectile dysfunction [1], abdominal pain [1], uroschesis [1]
160	Jatal S (166)	1L	28 y	Frequency, urgent & defecation pain

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