

CASE REPORT

Case Report: An occurrence of steinstrasse in retrograde intra renal surgery for a large staghorn kidney stone – difficulty managing surgical outcomes [version 1; peer review: 1 approved, 1 approved with reservations]

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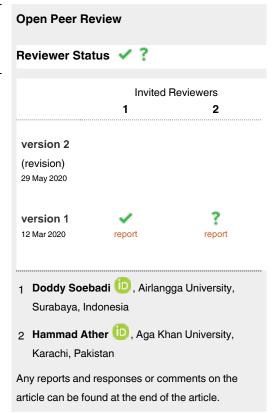
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Abstract

Immediate removal of staghorn stones is compulsory to prevent life-threatening complications. The advancement of endoscopic technology makes retrograde intrarenal surgery (RIRS) a favorable treatment to remove large stones over the standard percutaneous nephrolithotomy (PCNL). Without careful considerations, it can cause the formation of steinstrasse. Here, we present the case of a 68-year-old man with multiple stones along his right urogenital duct after being treated with RIRS to remove a staghorn stone. After 2 years of multiple interventions, the steinstrasse was completely removed. To prevent this complication, detailed assessment of the stone (size, location) and renal anatomy should be performed prior to the procedure.

Keywords

retrograde intrarenal surgery, staghorn stones, steinstrasse, complication





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Author roles: Birowo P: Conceptualization, Data Curation, Formal Analysis, Funding Acquisition, Investigation, Methodology, Project Administration, Resources, Supervision, Visualization, Writing – Review & Editing; **Rasyid N**: Conceptualization, Data Curation, Formal Analysis, Supervision; **Atmoko W**: Conceptualization, Data Curation, Formal Analysis, Methodology, Validation, Visualization; **Sutojo B**: Conceptualization, Visualization, Writing – Original Draft Preparation

Competing interests: No competing interests were disclosed.

Grant information: The author(s) declared that no grants were involved in supporting this work.

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Introduction

The term "staghorn" describes the configuration of large, branched renal stones that occupy the pelvis and extent to at least two renal calices. Immediate removal of the stones is compulsory to prevent serious kidney injury and life-threatening sepsis1. According to the American Urological Association, percutaneous nephrolithotomy (PCNL) is the standard treatment for staghorn removal². Recently, urologists started using retrograde intrarenal surgery (RIRS) to treat large stones since it is less invasive and simpler than PCNL³. However, this procedure might cause the formation of steinstrasse as a complication especially in stones of size 2-3 cm, which require more invasive removal procedures. This multi-procedure approach in the treatment of renal stone can impact the patients' quality of life, especially when the stone is hard (measured with Hounsfield unit of more than 1000)4,5. This study shows and discusses the formation of steinstrasse and the need of follow up procedures, which led to depression in the patient after the use of RIRS for staghorn stone removal.

Case presentation

A 68-year-old man came to our hospital in April 2016 with multiple stones along his right urogenital system. He had been experiencing flank pain that was not influenced by body position for 1 month. He denied any treatment relating to the pain that he experienced in this period. He also denied having a family history of this symptom nor ever he had this symptom before. Physical examination only revealed tenderness of the right flank.

Computed tomography (CT) urography at the previous hospital showed a staghorn stone at the right inferior calyx with a size of $45.7 \times 59.3 \times 27.5$ mm (Hounsfield unit not available), a left renal cyst with size of $19.2 \times 22.2 \times 18.9$ mm, and a third-grade right-side hydronephrosis (Figure 1). Post-RIRS photo showed a double J stent with multiple tiny stones from the right pelvio-calyces to vesico-ureteral junction (Figure 2a).

A month later at when he come to our hospital for a second opinion, his plain abdominal X-ray result had not changed

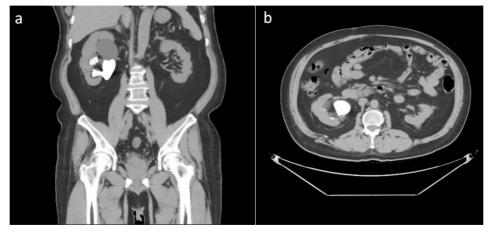


Figure 1. Initial computed tomography (CT) urography. The first CT Urography of the patient shows right staghorn stone with grade 3 hydronephrosis and left renal cyst.

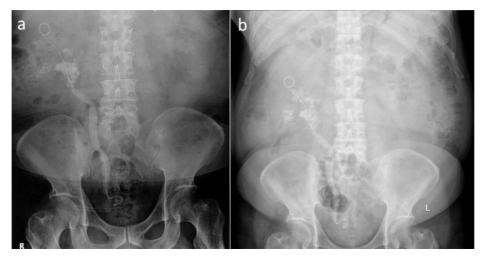


Figure 2. Steinstrasse formation. An immediate (a) and 1 month (April 2016) (b) Plain abdominal photo following retrograde intrarenal surgery shows multiple tiny stones along the right genitourinary system.

(Figure 2b). Right ureteroscopy (URS), right nephrostomy, and right PCNL was performed and post-operative X-ray imaging was conducted (Figure 3a). Another right URS was performed 2 weeks later, resulting in the remaining 8-mm stone at the ureter-pelvic junction (UPJ) (Figure 3b).

Extracorporeal shock wave lithotripsy (ESWL) was performed twice in June 2016, resulting in a decrease in stone size to 6 mm. (Figure 3c). Another ESWL was performed the next month (Figure 3d). In July 2016, he underwent a right laser URS followed by replacement of the DJ stent (Figure 3e). Three months later, another ESWL was performed (Figure 3f). Shortly after, the remaining DJ stent was removed. Plain abdominal X-ray still showed a residual right nephrolithiasis. (Figure 3g). In 2017, the patient was so depressed from the numerous procedures that he decided to end the treatment for his remaining stone. He admitted lacking spirit throughout the day since the failure of the last ESWL procedure and feeling that his stone would not be able to be cured, along with his continuous need for pain medication.

Almost two years later (January 2018), routine plain abdominal X-ray and CT urography showed no change on his right nephrolithiasis (Figure 3h, i). In June 2019, he was persuaded

by his family to re-try stone management and had the last RIRS at another hospital to remove the remaining stone completely (Figure 3j). In November 2019, he visited our hospital for DJ stent removal and plain abdominal X-ray. Neither stone nor DJ stent were observed. The summary of the patient's history of illness can be seen in Table 1.

Discussion

The management of nephrolithiasis has been changing dramatically over time, shifting from open surgery to less-invasive procedures, such as PCNL and ESWL⁶. According to the American Urological Association and European Association of Urology guidelines, the standard treatment for staghorn stone removal is PCNL^{2,5}. PCNL has a high stone-free rate (SFR), similar to that of an open surgery (93%). It also results in lower morbidity, shorter operative time, shorter hospital stays, and earlier back to work compared to open surgery. However, it can cause severe complications, such as renal trauma with severe uncontrollable bleeding^{7,8}.

On the other hand, the development of flexible ureteroscopes allows for excellent visualization that makes it a favorable procedure for most urologists. The possibility of using holmium

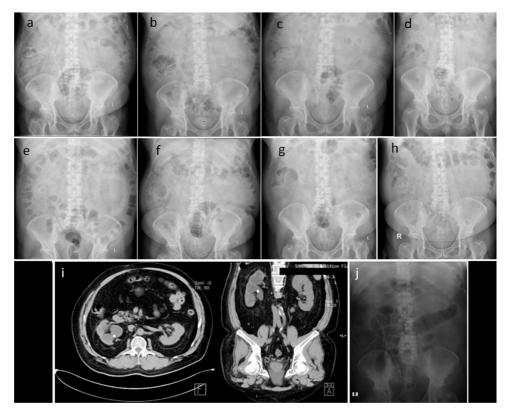


Figure 3. Sequential imaging photos. After right ureterorenoscopy, right nephrostomy, and right percutaneous nephrolithotomy in April 2016 (a), after ureterorenoscopy and percutaneous nephrolithotomy in June 2016 (b), after the second extracorporeal shock wave lithotripsy in June 2016 (c), after the third extracorporeal shock wave lithotripsy in July 2016 (d), after right laser ureterorenoscopy and replacement of right double J stent in July 2016 (e), after extracorporeal shock wave lithotripsy in October 2016 (f), double J stent removal in October 2016 (g), routine control in January 2018 (h & i), retrograde intrarenal surgery which shows no residual stone in June 2019 (j).

Table 1. Summary of the patient's history of illness.

Time	Initial condition	Procedure	Result
December 2015	Plain abdominal photo showed right staghorn stone (45,7 \times 59,3 \times 27,5 mm); Third grade hydronephrosis	RIRS and DJ stent insertion	Multiple tiny stones along the right genitourinary system from pelvio-calyces to vesico-ureteral junction (Figure 2)
April 2016	Plain abdominal photo showed multiple tiny stones along the right genitourinary system from pelvio-calyces to vesico- ureteral junction	Right URS; Right nephrostomy; Right PCNL; Insertion of a new DJ stent	A remaining radio opaque stone with a diameter of 8 mm at the UPJ (Figure 3a)
June 2016	Plain abdominal photo showed a radio opaque stone with the size of 8 mm	ESWL twice	The stone size was decreased to 6 mm (Figure 3b, c)
July 2016	Plain abdominal photo showed a radio opaque stone with the size of 6 mm	ESWL; DJ stent replacement; Right laser URS	Small residual stones at the right kidney (Figure 3d, e)
October 2016	A residual right nephrolithiasis	ESWL; DJ stent removal	A residual right nephrolithiasis (Figure 3f)
January 2018	Plain abdominal photo and CT urography showed right nephrolithiasis	N/A	Figure 3g
June 2019	CT urography showed right nephrolithiasis	RIRS and DJ stent insertion	Right DJ stent <i>in situ</i> ; No residual stone (Figure 3h, i)
November 2019	Right DJ stent in situ; No residual stone	DJ stent removal	No stone was found on the final BNO (Figure 3j)

RIRS, retrograde intrarenal surgery; URS, Ureteroscopy; PCNL, percutaneous nephrolithotomy; ESWL, extracorporeal shock wave lithotripsy.

lasers alongside a ureteroscope, lowering the cost, has made this procedure even more popular⁹. Compared to PCNL, RIRS has a slightly lower SFR of 87% and also lower morbidity and complication rate at 2%¹⁰. In our case, the first option of RIRS instead of PCNL was because the patient preferred a less invasive method.

In Indonesia, PCNL is still the first choice for treating large renal calculi according to *Ikatan Ahli Urologi Indonesia* (the Indonesian Urologist Association). However, the use of PCNL in Indonesia is still limited due to the lack of technology and experts, particularly in remote areas¹¹. The incidence of stein-strasse formation after RIRS is 20% of large renal stone cases followed by hydronephrosis⁴. The development of SS is also seen in our patient who were initially treated with RIRS. To avoid this complication, a scoring system was developed by Resorlu *et al.*¹² that includes four indicators: a renal stone size >20 mm, lower pole stone with an infundibulum-pelvic angle <45°, a stone number in different calyces >1, and abnormal renal anatomy. A greater score is associated with a lower SFR. This score can be calculated prior to RIRS.

Urolithiasis is a painful chronic disease which significantly impacts the quality of life of the sufferers. In addition to chronic pain, the acute pain of urolithiasis resulting from stone movement often causes fear of recurrence. Recent studies have suggested an association between the disease and anxiety and depression. In the present study, our patient developed symptoms of depression during the second year of his treatment because he had to undergo multiple surgical procedures in a year. However, after receiving support from his family and reassurance from the clinicians, the patient finally decided to continue treatment for his leftover stones. The patient initially preferred RIRS to PCNL

because it was less invasive, but after his negative experiences, he would prefer PCNL to RIRS.

RIRS may be used in cases where open surgery and ESWL are risky or inadequate, such as in patients with obesity, bleeding disorders, musculoskeletal deformities, renoureteral malformations, and infundibular stenosis⁵.

One limitation of this study is that we did not know the hardness (Hounsfield units) of the stone prior to the patient presenting to our clinic, meaning we could not specifically determine the cause of his previous failure of treatment other than the size of the stone.

Conclusions

RIRS can be an alternative choice to remove large staghorn calculi in selected patients even though the efficacy is lower than PCNL. Detailed assessment should be made to prevent any complications. Patients with large staghorn calculi should be informed regarding the possible need for multiple procedures to completely remove the stones, otherwise receiving inadequate information will lead the patient to negative thinking and treatment avoidance.

Data availability

All data underlying the result are available as part of the article and no additional source data are required.

Consen

Written informed consent for publication of their clinical details and/or clinical images was obtained from the patient/parent/ guardian/relative of the patient.

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Reviewer Report 04 May 2020

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? Hammad Ather 📵

Department of Surgery, Aga Khan University, Karachi, Pakistan

- Steinstrasse is a legacy of SWL (Does ureteral stenting prior to shock wave lithotripsy influence the need for intervention in steinstrasse and related complications? (Ather et al., 2009¹)) and RIRS for larger stones is no different from performing SWL under endoscopy. The authors should discuss this point.
- 2. Conclusions are contrary to the case account. The conclusions should be that larger stones (Staghorn) would preferably not be treated by RIRS.
- 3. Few minor issues:
- In the abstract I don't agree with the word "compulsory" a more preferable alternative would be "desirable" or "important" etc.
- RIRS is not mentioned as a treatment option for large kidney stones.
- Mention other complications of the use of RIRS besides steinstrasse for large renal stones.
- The phrase "RIRS to remove a staghorn stone" in the abstract is not an appropriate reflection of the abilities of RIRS in that set up.
- There are many grammatical errors that need attention.
- The authors used the phrase "which led to depression in the patient after the use of RIRS for staghorn stone removal". This needs to be rephrased for clarity as to what the authors are implying. Are they saying prolonged treatment causes psychological issues, like depression? Please cite a reference.
- In the case presentation, the authors write "multiple stones along his right urogenital system" this needs to be rewritten for clarity. Stones were in the collecting system of the right kidney.



- With the limited access to single section I disagree with the authors' interpretation of the CT. I see a Staghorn stone with a major component in the renal pelvis and branching calculi in lower and middle pole calyces. There is dilatation and obstruction with ballooning of the upper pole calyx due obstruction of the infundibulum of the upper pole calyx. There is no cyst visible in these scans.
- It is grade three, not third grade hydronephrosis, which is incidentally also not clear in the CT cut shown.
- The authors should avoid using term "genitourinary".
- What is the stone composition?

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Is the background of the case's history and progression described in sufficient detail?

Are enough details provided of any physical examination and diagnostic tests, treatment given and outcomes?

Yes

Is sufficient discussion included of the importance of the findings and their relevance to future understanding of disease processes, diagnosis or treatment?

Yes

Is the case presented with sufficient detail to be useful for other practitioners? Partly

Competing Interests: No competing interests were disclosed.

Reviewer Expertise: Urolithiasis and Bladder cancer

I confirm that I have read this submission and believe that I have an appropriate level of expertise to confirm that it is of an acceptable scientific standard, however I have significant reservations, as outlined above.

Author Response 23 May 2020

Ponco Birowo, Faculty of Medicine Universitas Indonesia - Cipto Mangunkusumo Hospital, Jakarta Pusat, Indonesia

Comments from Reviewer and Author Response:

Steinstrasse is a legacy of SWL (Does ureteral stenting prior to shock wave lithotripsy
influence the need for intervention in steinstrasse and related complications? (Ather et al.,



2009¹) and RIRS for larger stones is no different from performing SWL under endoscopy. The authors should discuss this point.

Author response:

We have revised and added this additional paragraph to our manuscript:

The necessity for routine stent insertion before or after RIRS to increase stone clearance remains unclear. The main purpose of stent insertion is to prevent ureteral stricture, accelerate healing, and facilitate stone passing (Cleynenbreugel, et al., 2017). On the other hand, stent insertion increases the possibility of urinary tract infection, dysuria, pollakiuria, hematuria, and repeated cystoscopy may be required in cases of stent migration and to assess the need for extraction (Ozyuvali, et al., 2015). Stent insertion before shockwave lithotripsy (SWL) does not eliminate the need for intervention in the management of steinstrasse (Ather et al., 2009). In the present case, considering the size and the position of the stone, ureteral stent placement prior to RIRS would be difficult and other options should be considered.

Conclusions are contrary to the case account. The conclusions should be that larger stones (Staghorn) would preferably not be treated by RIRS.

Author response:

Thanks for your advice. We have revised our conclusion to become: RIRS is not the preferable option to remove large staghorn calculi due to lower efficacy and other possible complications. It can be used in circumstances where open surgery or PCNL is not possible, but careful assessment is necessary to determine whether the procedure will be beneficial and safe for the patient.

• In the abstract I don't agree with the word "compulsory" - a more preferable alternative would be "desirable" or "important" etc.

Author response:

Thank you for your advice, we have changed the word according to your advice.

RIRS is not mentioned as a treatment option for large kidney stones.

Author response:

We have revised and added this additional paragraph to our manuscript: Initially, the use of RIRS is limited to patients who cannot undergo PCNL or shockwave lithotripsy (SWL) due to several contraindications. However, with the development of technology, the usage of RIRS for large stone is now possible. Compared to PCNL, RIRS has a slightly lower SFR of 87% and also lower morbidity and complication rate of 2%. ¹⁰ In our case, the use of RIRS instead of PCNL as the first treatment was due to the patient's preference for a less invasive method.

Mention other complications of the use of RIRS besides steinstrasse for large renal stones.

Author response:

We have revised and added this additional paragraph to our manuscript: RIRS is a less-invasive procedure compared to PCNL. Complications may arise intra- or post-operatively in some cases but are usually minor and manageable. The common complications of RIRS include hemorrhage, intrapelvic hematoma, mucosal injury, ureteral perforation and avulsion, urinary tract infection, and sepsis. In a study by Niwa et al., the most



common complication associated with RIRS in treating staghorn stones was urinary tract infection (Clavien-Dindo II, 28.2%), followed by fever (7.7%), general malaise (2.6%), and malposition of a ureteral stent (2.6%).

• The phrase "RIRS to remove a staghorn stone" in the abstract is not an appropriate reflection of the abilities of RIRS in that set up.

Author response:

We have revised our abstract as it is not aligned with the insight of our case presentation that do not recommend the use of RIRS for large stones removal. Here is the revised version: Immediate removal of staghorn kidney stones is important to prevent life-threatening complications. With the advancement of endoscopic technology, retrograde intrarenal surgery (RIRS) is now an alternate treatment to the standard percutaneous nephrolithotomy (PCNL) for stones removal. However, when used to treat large stones (>3cm), RIRS can cause the formation steinstrasse (SS).

There are many grammatical errors that need attention.

Author response:

Thank you for addressing your concern. We have revised the grammatical error that we found.

• The authors used the phrase "which led to depression in the patient after the use of RIRS for staghorn stone removal". This needs to be rephrased for clarity as to what the authors are implying. Are they saying prolonged treatment causes psychological issues, like depression? Please cite a reference.

Author response:

We have added some information to clarify this issue.

Urolithiasis is a painful chronic disease that has significant impacts on a patient's quality of life. In addition to chronic pain, the acute pain of urolithiasis resulting from stone movement often causes fear of recurrence. Recent studies have suggested an association between the disease and anxiety and depression. In the present study, our patient developed symptoms of depression during the second year of his treatment because he had to undergo multiple surgical procedures within a year to remove the SS. In addition, the patient had to endure the pain associated with recovery after each procedure, as well as the pain caused by the remaining stones.

In the case presentation, the authors write "multiple stones along his right urogenital system"
 this needs to be rewritten for clarity. Stones were in the collecting system of the right kidney.

Author response:

Thank you for your correction. We have updated the term accordingly.

With the limited access to a single section I disagree with the authors' interpretation of the CT. I see a Staghorn stone with a major component in the renal pelvis and branching calculi in lower and middle pole calyces. There is dilatation and obstruction with ballooning of the upper pole calyx due to obstruction of the infundibulum of the upper pole calyx. There is no cyst visible in these scans.



Author response:

Thank you for your concern. We have decided to change the picture:

• It is grade three, not third-grade hydronephrosis, which is incidentally also not clear in the CT cut shown.

Author response:

We have changed "third grade" to "grade three" hydronephrosis.

The authors should avoid using term "genitourinary".

Author response:

Thank you. We have altered "genitourinary" to urinary system.

What is the stone composition?

Author response:

Unfortunately, in the present case, the stone composition was not analyzed due to financial constraints.

 This is a good example of how minimally invasive surgeries can take a long journey before they come to a satisfactory result.

Author response:

Thank you for your appreciation.

• The authors did not mention the drawbacks of these lengthy managements, i.e. the opinion from the patient side and the cost of the serial treatments.

Author response:

The patient self-funded the whole treatment but he refused to tell the amount he had to pay. As for his opinion about the lengthy procedure, the patient complained of slight depression due to the continuous pain, either from the remaining stone and from the procedures. This information is mentioned in the case presentation and discussion section.

 But this is a very good clinical experience to be one of the considerations before the urologist(s) offer this minimally invasive procedure(s).

Author response:

Thank you. We hope this study provides a handful of insight regarding the disadvantage of RIRS to treat large kidney stone.

Competing Interests: No competing interests were disclosed.

Reviewer Report 14 April 2020



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Doddy Soebadi (10)

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- This is a good example of how minimally invasive surgeries can take a long journey before they come to a satisfactory result.
- The authors did not mention the drawbacks of these lengthy managements, i.e. the opinion from the patient side and the cost of the serial treatments.
- But this is a very good clinical experience to be one of the considerations before the urologist(s) offer this minimally invasive procedure(s).

Is the background of the case's history and progression described in sufficient detail? Yes

Are enough details provided of any physical examination and diagnostic tests, treatment given and outcomes?

Yes

Is sufficient discussion included of the importance of the findings and their relevance to future understanding of disease processes, diagnosis or treatment?

Yes

Is the case presented with sufficient detail to be useful for other practitioners? Yes

Competing Interests: No competing interests were disclosed.

Reviewer Expertise: General urology, voiding dysfunction, sexual medicine, endourology, laser urology.

I confirm that I have read this submission and believe that I have an appropriate level of expertise to confirm that it is of an acceptable scientific standard.

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