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STONES/ENDOUROLOGY

REVIEW

# Familial prostatic calcification in childhood associated with cranial-bone thickening: Review of literature and report of three cases

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**KEYWORDS**

Childhood;
Prostatic calculi;
Cranial bone

**Abstract** ***Objectives:*** To review the few published cases of prostatic calculi, a rare condition in children, and to report three further cases.

***Methods:*** The databases PUBMED and HINARI were searched using the keywords 'childhood' and 'prostatic calculi'; the search included reports from 1956 to the present. Further cases from three families with children having prostatic calculi are reported here.

***Results:*** Four cases were recorded previously but no association was stated between the presence of calculi and cranial-bone abnormality.

***Conclusions:*** Prostatic calculi in childhood are rare; the condition requires further study and clarification.



## Introduction

Prostatic calculi are rare in children, infrequent in those aged $<40$ years and common in men aged $>50$ years [1]. Reports of paediatric cases are very scarce [2]. A check of previous reports suggests an increased incidence of urinary calculi in ochranosis [3]. No specific single factor has been identified as responsible. Here we present a review of previous cases and report three more patients.

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## Methods

The databases of PUBMED and HINARI were searched using the keywords 'childhood' and 'prostatic calculi', the search extending from 1956 to the present. The review included 16 articles but only four contained records related to children. After obtaining approval from the ethics committee for the study, and parents' approval for publishing the photographs, we present further cases from three families who have children with prostatic calculi.

## Results

The articles published in the 1950s contained no abstract; Table 1 shows a summary of the review. In the article by Schonlebe [4] the number of cases was not given. Spence and Chenoweth [5] reported two cases; they were Müllerian ducts cysts containing calculi. Izzidien [2] reported the first case in Iraq in 1980. Uchijima et al. [6] reported in 1984 an infant with stones of the seminal vesicles and ejaculatory duct.

### *Case 1: The first family*

A 5-year-old (Iraqi) boy was first seen in 1978 (Fig. 1); he had been to another hospital, where he had an open vesicolithotomy. Physical examination revealed a fair-haired healthy boy, and a DRE was unremarkable. IVU showed obvious prostatic calcification but normal kidneys and bladder (Fig. 2). He had no contact for several years and obviously had several operations for vesical stones; finally he had a urinary diversion (ileal conduit). When he attended in 1999 he was blind, and a plain film showed that the calcification had grown to a huge size. Stone analysis from previous operations revealed mixed Whewellite (calcium oxalate), Brushite (acid calcium phosphate) and ammonium acid urate, with no prevalent type. Ophthalmological examination revealed bilateral optic nerve atrophy.

The family was asked to bring his only brother, who was 7 years old (Fig. 3); he was asymptomatic but a plain film showed prostatic calcification. Both brothers were investigated, including IVU, a complete blood analysis, erythrocyte sedimentation rate, and measurements of blood urea, creatinine, calcium, phosphate, uric acid, parathyroid hormone, and alkaline phosphatase (Table 2). In addition 24-h urinary calcium, uric acid, cystine and oxalate were analysed. All these investigations showed no obvious abnormality. Cysto-urethroscopy showed no bladder or urethral pathology. Both brothers were followed. No specific treatment was prescribed.

### *Case 2: The second family*

A 4-year-old (Iraqi) boy was seen in 2000 (Fig. 4); he had been to another hospital where an open vesicolithotomy was performed and a suprapubic cystostomy catheter inserted. Physical examination revealed a healthy boy with a suprapubic catheter; a DRE was unremarkable. Cysto-urethroscopy (Fig. 5) showed stones obstructing the prostatic urethra, some of which were protruding from prostatic recesses. These small diverticulae were within the prostate and distal to the bladder neck. The bladder was opened and the stones were evacuated, taking care not to open further prostatic tissues or incise directly on palpable stones. He had an uneventful postoperative course. Later the suprapubic catheter was removed and he could void normally. Stone analysis showed a mixed type with no prevalent item.

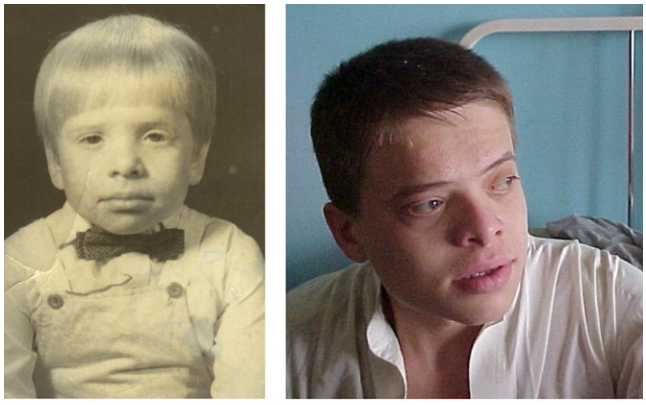


**Figure 1** First family: The second photograph was taken when the boy became totally blind.

The family was asked to bring his 6-year-old brother (Fig. 6), the only male sibling, and he was asymptomatic. His plain film showed prostatic calcification. Both brothers had full haematological and biochemical investigations, and none showed any abnormality (Table 2). The IVU and cysto-urethroscopy were normal. The patient and his brother were lost to follow-up.

### *Case 3: The third family*

A 4-year-old (Iraqi) boy was seen in 2000 (Fig. 7); he was also treated elsewhere by open vesicolithotomy with insertion of suprapubic cystostomy catheter. A plain film showed prostatic calcification. It showed also late changes of congenital dislocation of the right hip joint. CT of the patient showed a diffuse thickening of the skull, base and calvarium (the upper domed portion of the skull), involving both inner and outer tables and diploid space. There was narrowing of both of the optic foramens due to over-pneumotised ethmoidal air cells and thickening of the skull base. There was evidence of triventricular hydrocephaly (Fig. 8).

**Table 1** A summary of the previous reports.

| Reference | No. of cases | Year | Aetiology | Diagnosis | Treatment |
|---|---|---|---|---|---|
| [4] | ? | 1956 | Unknown | Prostatic calculi | ? |
| [5] | 2 | 1957 | Unknown | Müllerian duct cyst/calculi | Open surgery |
| [2] | 1 | 1980 | Unknown | Prostatic calcification and ureteric stone | Open surgery for the ureteric stone only |
| [6] | 1 | 1984 | Unknown | Stones in seminal vesicles and ejaculatory duct | Open surgery |
| Present | 3 | 2011 | Unknown | Prostatic calculi | Open surgery |

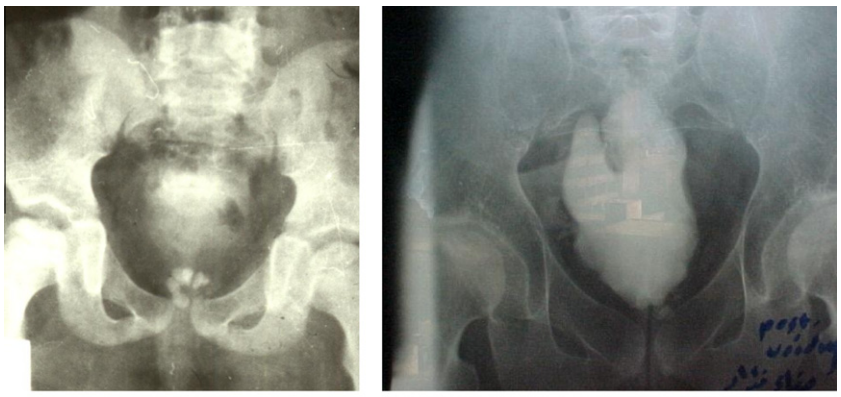


**Figure 2** A plain X-ray of the pelvis showing the increase in stone size.

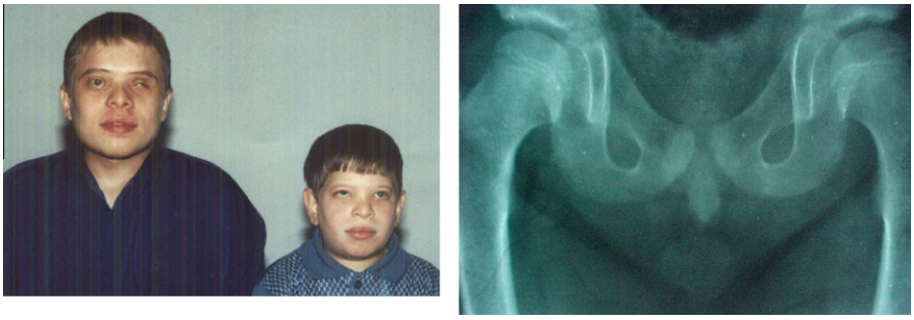

**Figure 3** A photograph of the brother, and his pelvic plain X-ray.

**Table 2** The biochemistry results.

| Source, analyte | Family | | | | | |
|---|---|---|---|---|---|---|
| | 1st | | 2nd | | 3rd | |
| | Patient | Brother | Patient | Brother | Patient | Sister |
| *Blood* | | | | | | |
| Urea (mg/dL) | 30 | 25 | 33 | 33 | 28 | 37 |
| Creatinine (mg/dL) | 1.0 | 1.1 | 0.9 | 0.8 | 0.9 | NA |
| Calcium (mg/dL) | 9 | 8.5 | 9.2 | NA | 8 | 8.2 |
| Phosphate (mg/dL) | 4 | 4.5 | 4 | 3.8 | 4.7 | 4.8 |
| Uric acid (mg/dL) | 6 | 5 | 4.5 | 4 | 5.5 | 4 |
| Alkaline phosphatase (IU) | 5 | 5 | 6.2 | 4 | 3.5 | 4.2 |
| Parathyroid hormone (ng/L) | 30 | 45 | 32 | 42 | 33 | 23 |
| *Urine, 24 h* | | | | | | |
| Calcium (mg) | 253 | 200 | 270 | 155 | 160 | 160 |
| Uric acid (mg) | 200 | 150 | 220 | 150 | 105 | 100 |
| Cystine (mg) | 22 | 20 | 18 | NA | 10 | 17 |
| Oxalate (mg) | 20 | 24 | 18 | 21 | 17 | 16 |

NA, not available.

After asking about other male members of the family, they reported no male sibling, but there was a 13-year-old blind sister. Her skull CT (Fig. 9) showed marked thickening of the cranial bones (vault, base, etc.). On examination she had bilateral optic nerve atrophy. The boy was discharged after the suprapubic catheter was removed. The family was asked to attend for follow-up.

## Discussion

Prostatic calculi have been described in cases of ochronosis, and our review suggested an increased incidence of urinary calculi in this syndrome [3]. Prostatic calcification in the adult is not uncommon [7]. The seminal vesicle or the utricle can be the site of calcification or stone formation [8]. Small, multiple

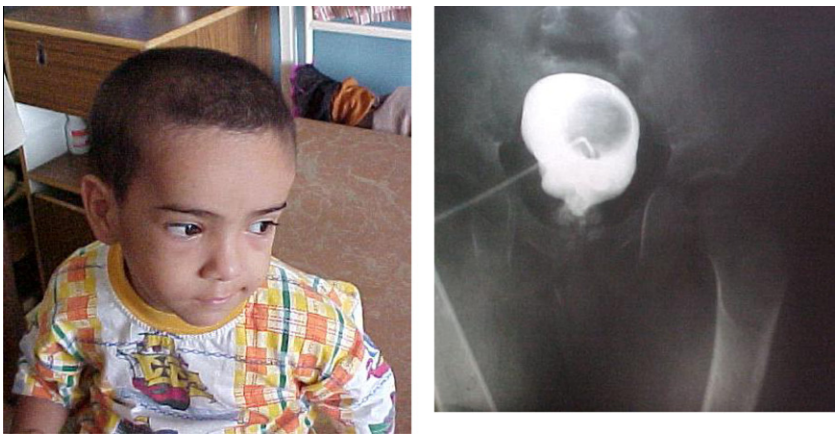

**Figure 4** Second family: A cystogram shows the suprapubic catheter and prostatic calculi.

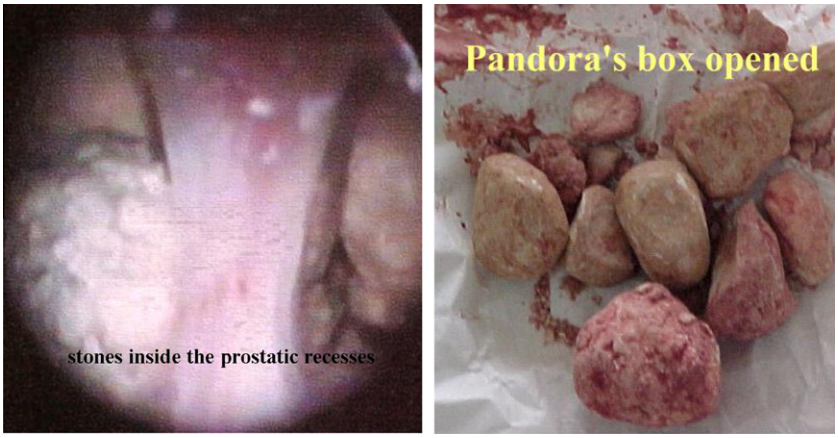


**Figure 5** Stones seen inside the prostatic recesses and after extraction.

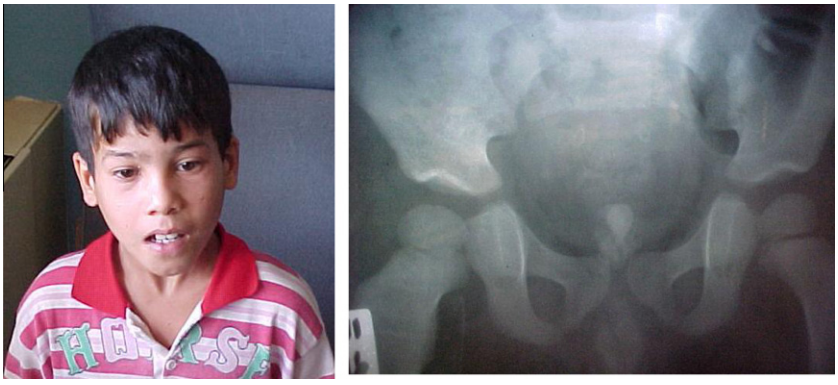

**Figure 6** The asymptomatic brother, with his pelvic plain X-ray

calcifications are a normal, often incidental ultrasonographic finding in the prostate, and represent a result of age rather than a pathological entity. However, larger prostatic calculi might be related to underlying inflammation and require further evaluation, and possibly treatment [9]. The presence or volume of prostatic calculi had no significant effect on serum PSA levels. Results suggested that the influence of prostatic calculi is irrelevant in men with elevated PSA levels [10].

All the present patients were primarily seen by general surgeons in the provinces. They were explored without endoscopic examination. No stones were found in the bladder. They had undergone unnecessary open surgery, which had led to further stone formation that became vesical rather than prostatic. Once the prostate is incised from the bladder, stones continue to be formed and discharged into the urethra and bladder; this requires further operations. This situation is like Pandora's box.

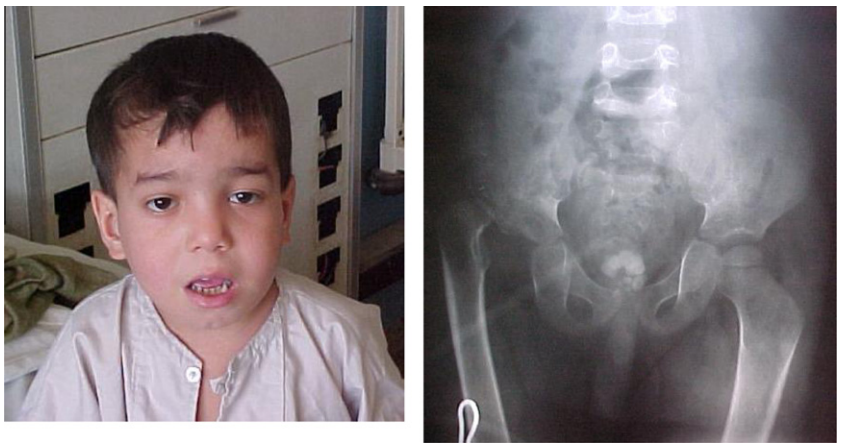

**Figure 7** Third family: A photograph and plain X-ray of the patient's abdomen.

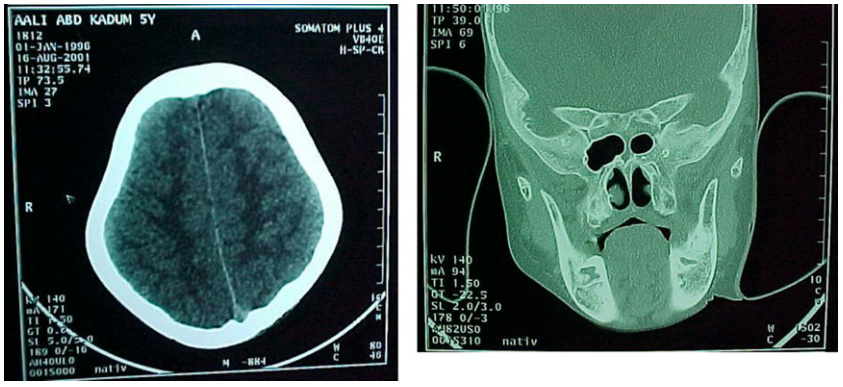


**Figure 8** CT showing diffuse thickening of the skull, base and calvarium, involving both inner and outer tables and diploid space. There was narrowing of both of the optic foramens due to over-pneumotised ethmoidal air cells and thickening of skull base. There was evidence of triventricular hydrocephaly.

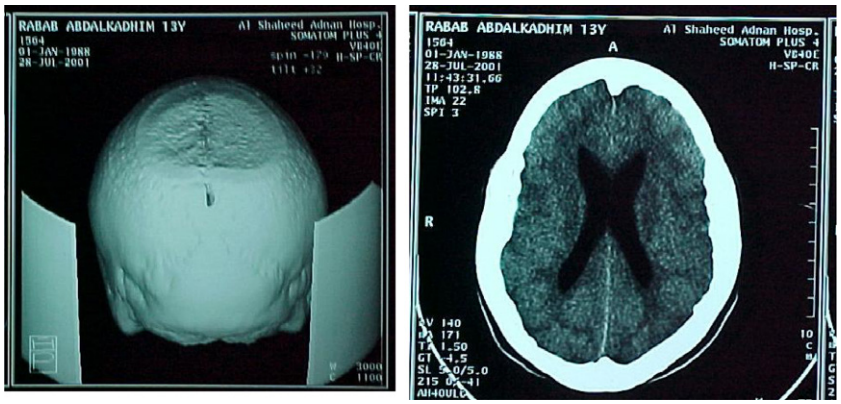


**Figure 9** Skull CT of the patient's 13-year-old sister, showing marked thickening of the cranial bones (vault, base, etc.). She had bilateral optic nerve atrophy.

Calcium was considered as a possible factor. It was found that calcium and the urinary calcium/creatinine ratio was significantly increased in children with all types of urinary symptoms. Fallahzadeh et al. [11] recommended that urinary calcium should be measured in all children with urinary tract symptoms, especially if unexplained. Calcium levels were not elevated in the present cases.

Studies of prostatic calculi in childhood are rare. Reports linking prostatic calcification to either cranial-bone thickening or intracranial calcification are even rarer. Some authors studied the counteracting functions between the pineal gland and prostate. Is there a relation between pineal concretions and prostatic calculi? A low correlation was reported [12] and that study recommended the inclusion of younger patients.

The other element was cranial-bone thickening with optic nerve atrophy as a correlation. Skull involvement and blindness was obvious in the present cases. These rare pathologies were found repeatedly. Siblings had the same pathology, and a female sibling also had skull involvement and blindness. We report for the first time these associated phenomena justify further verifications and clarifications.

Further investigations are mandatory to explore the genetic basis of prostatic calcification in children, and its association with cranial-bone thickening and optic nerve atrophy. Collaboration is needed between urologists and molecular geneticists. Thus, it would be useful to design a worldwide multicentre study. Accumulating further cases will certainly help in authenticating this special situation in clinical urology practice. Are we dealing with a new medical rarity? Is it a new syndrome?

## Conflict of Interest

The authors have no conflict of interest to declare.

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