

Asbestos exposure and malignant mesothelioma of the tunica vaginalis testis: Case series and review of the literature

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Abstract

Introduction

We aim to report a series of cases of asbestos exposure and malignant mesothelioma of the tunica vaginalis.

Materials and methods

Nine men diagnosed with malignant mesothelioma of the tunica vaginalis testis sought legal representation with law firms around the United States, including the principal author's firm. Fewer than 230 cases of mesothelioma of the tunica vaginalis have been reported in the worldwide literature, with documented asbestos exposure rates ranging from 0% to 70%.

Results

The men in this case series had a variety of exposures to asbestos. Many were exposed as children during home renovation work or to dust brought by relatives to their homes. Adult exposures included those from home renovation and construction work, vehicle maintenance, shipyard work and exposures at railroads. The pattern of exposures is similar to that described by Welch and colleagues in a case series of men with peritoneal mesothelioma and by Boffetta in a review of peritoneal mesothelioma in cohort studies.

Conclusion

Malignant mesothelioma of the tunica vaginalis testis is a variant of

peritoneal mesothelioma and shares similar asbestos causation.

Introduction

During normal human embryonic development, the testes descend from the abdominal cavity to the scrotal sac. Passage occurs through the processus vaginalis, which arises as an outpouching of the parietal peritoneum at the beginning of the third month of gestation. After the testis descends into the scrotum (between 7 and 9 months of gestation), the processus vaginalis is normally obliterated. However, the processus vaginalis remains patent at birth in 20% of the population. Most boys born with a patent processus vaginalis remain asymptomatic throughout life, but incomplete closure of the processus vaginalis may lead to a variety of abnormalities. Complete patency may result in a communicating hydrocele or a congenital inguinoscrotal hernia.

A persistent processus vaginalis often closes during the first year of life, probably in response to the surge in serum testosterone that normally occurs at 1–2 months of age¹. The tunica vaginalis is a mesothelium-lined sac that results from closure of the superior portion of the processus vaginalis. In the event that the processus vaginalis does not close completely, then the tunica vaginalis remains in communication with the peritoneal cavity.

Mesothelioma of the tunica vaginalis is a rare tumour. In a study of incident cases of mesothelioma in the Netherlands, most of the mesotheliomas occurred in the pleura, where there were 119 (88%) against 15 (11%) in the peritoneum and

two in the tunica vaginalis testis². Marinaccio and colleagues³ reported on the incidence of extrapleural mesothelioma in the Italian National Mesothelioma Registry. Standardized incidence rates (Italy, 2004, per million inhabitants) were 2.1 cases for the peritoneal site and 0.2 cases for the tunica vaginalis testis. Mesothelioma of the tunica vaginalis represents only 0.09% (10 cases) of all mesothelioma deaths in the UK Health and Safety Executive Mesothelioma Register⁴.

Mesothelioma of the tunica vaginalis has been associated with asbestos exposure. The first case report was published by Fligel and Kaneko in 1976⁵. The subject, a 68-year-old man, had worked for 40 years as a pipe insulator. Gorini and colleagues⁶ in Italy reported two cases of this tumour. A 67-year-old man had been occupationally exposed to asbestos for a 30-year period with a latency of 42 years. An 80-year-old man had been exposed to asbestos for 5 years with a latency of 52 years. Spiess and colleagues reported a retrospective clinicopathological and follow-up study of five patients with malignant mesothelioma of the tunica vaginalis treated at the M.D. Anderson Cancer Center in Houston, Texas, during a 25-year period⁷. Asbestos exposure was identified in four of the five patients. In the Italian Registry Study³, 70% of subjects with mesothelioma of the tunica vaginalis had occupational or leisure-related exposure to asbestos. Mean latency (defined as the time elapsing between the beginning of exposure to asbestos and diagnosis) was estimated for cases with sufficient information and was 46.8 years

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for the 13 cases with mesothelioma of the tunica vaginalis. With respect to industrial sector, the incidence rates were 0.24 cases per million person-years in construction, 0.25 cases per million person-years in the metal and steel mechanical industry, 0.36 cases per million person-years in the textile sector and 0.37 cases per million person-years in 'Transport and construction, transport maintenance and repair'.

We report here the asbestos exposure histories of nine American men with the diagnosis of mesothelioma of the tunica vaginalis testis who sought legal representation with law firms in the United States. The purpose of this report is to illustrate the variety of exposures, which led to the development of disease in these individuals.

Materials and methods

Nine men diagnosed with malignant mesothelioma of the tunica vaginalis testis sought legal representation with law firms in the United States. Their personal, medical and work histories, and information about their asbestos exposures, were developed in the usual course of litigation through investigation, record collection, deposition testimony, written discovery and consultation with expert witnesses. This information was collected by the subjects' various lawyers during their legal cases and was provided, with express written permission, to the principal author who sought it out and collected it for purposes of this report.

Results

Case 1:

Case 1 is a Caucasian male aged 60 years at the time of diagnosis. Malignant mesothelioma was diagnosed on the right side of the scrotum after surgical biopsy and immunohistochemical staining. Surgical resection of the tumour, testicle, spermatic cord, tunica vaginalis, tunica albuginea and epididymis

was performed. The subject died 15 months after diagnosis following tumour recurrence.

The subject was first exposed to asbestos around the age of 8 or 9 (53 years prior to diagnosis) years when his father removed asbestos insulation from the heating pipes in the basement of the family home. The asbestos fibre type in that insulation is unknown. In the mid- to late-1970s, he had exposure to chrysotile-containing products such as joint compound, furnace cement, flooring and roofing materials while doing renovation and repair work on his homes. While working as a civil engineer for a road contractor, in the 1970s, he had frequent exposure to dusts from crocidolite- and chrysotile-containing asbestos-cement pipe used for sewers and water lines. Some of this exposure was as a supervisor/bystander and some was hands-on cutting and grinding the pipe with power tools. He also had occasional bystander exposure at his employer's garage to friction products and gaskets (likely chrysotile-containing) used with the construction equipment during routine maintenance and repair work.

Case 2:

Case 2 is a Caucasian male aged 70 years at the time of diagnosis. Malignant mesothelioma (epithelioid type) was diagnosed on the right side of the scrotum after surgical biopsy and immunohistochemical staining. The tumour involved the tunica vaginalis and tunica albuginea and invaded into the testis and lymphatics. Surgery was performed 5 weeks after diagnosis, including resection of the right scrotal sac, testis, epididymis and vas deferens. He was still alive 46 months after diagnosis with no known recurrence.

He was first exposed to asbestos as a young child (64 years prior to diagnosis) through household contact with his father and intermittent contact with an uncle, both of whom

worked at a local shipyard during World War II. The asbestos fibre type from any potential exposure through the shipyard is unknown but is likely to have been mixed. He had sporadic paraoccupational exposure to asbestos through work on home renovation projects in the late 1950s and 1970s that included the use of chrysotile-containing joint compound, flooring material and home siding. Occupationally, he worked as a professional vehicle and equipment mechanic for 30 years from the 1950s to the 1980s including some time as a professional truck driver. He also performed maintenance work on family automobiles and farm equipment. These automotive and equipment exposures included chrysotile-containing friction materials, chrysotile-containing gaskets, chrysotile-containing automotive undercoatings and amphibole-containing (crocidolite and anthophyllite) automatic transmission parts. Many vintage samples of these products were recovered from the workshop at his home. Every chrysotile product that was tested revealed some level (all <1%) of amphibole contamination (mostly tremolite/actinolite with occasional amosite or 'other amphibole'). Crocidolite and anthophyllite in the automatic transmission parts was confirmed by light microscopy and transmission electron microscopy. In the case of the crocidolite parts, crocidolite was also confirmed by the product manufacturer's material safety data sheets.

Case 3:

Case 3 is a Caucasian male aged 59 years at the time of diagnosis. He sought treatment for a right-sided testicular mass (present for approximately 5 years prior), and a diagnosis of malignant mesothelioma of the tunica vaginalis testis (epithelioid type) was made by surgical biopsy. Computed tomography scans of his chest revealed bilateral pleural plaques and possible evidence of

fibrosis. He was still alive 71 months after diagnosis.

He was first exposed to asbestos 48 years prior to his diagnosis when he began to help his father with building renovation work involving the use of chrysotile-containing drywall taping compounds and floor tiles, and potentially other products of mixed fibre types used in the renovation work. He was later exposed to asbestos from chrysotile-containing drywall taping compounds and floor tiles while helping a family member on residential electrical jobs. He served in the United States Navy for 4 years and repaired aircraft turbines during two of those years while stationed aboard an aircraft carrier (mixed fibre types). He also worked many years for the railroad where he had additional exposures to insulation and other materials (mixed fibre types).

Case 4:

Case 4 is a Caucasian male aged 44 years at the time of diagnosis. Malignant mesothelioma (epithelioid type) of the spermatic cord was diagnosed on the right side of the scrotum by surgical biopsy and immunohistochemical staining. Surgical resection of the tumour, right testicle and spermatic cord was performed less than 1 month after diagnosis. The tumour recurred repeatedly over the course of the next year, including on the left side, and required additional surgeries and biopsies. Each subsequent biopsy confirmed the original diagnosis of malignant mesothelioma. Disease progression continued, and he died later that year at the age of 45 (14 months after diagnosis) years.

He was first exposed to asbestos 26 years prior to his diagnosis when he began work as a 'scrapper' and equipment operator for a mining company (fibre type unknown). He worked for 24 years in the service department at an auto-dealership as a mechanic and later as a service consultant. He was exposed to asbestos

from his own work on brakes and from the work of others working with brakes (most likely chrysotile with probable trace levels of tremolite/actinolite).

Case 5:

Case 5 is a Hispanic male aged 74 years at the time of diagnosis. Malignant mesothelioma was diagnosed on the right side after an 'inguinal canal mass' was discovered during surgery for an elective hernia repair. Pathological examination and additional immunohistochemical staining confirmed the diagnosis of mesothelioma originating in the tunica vaginalis testis. A right orchiectomy was performed a few months after diagnosis. Chest radiographs were suggestive of bilateral pleural thickening and pleural asbestosis. Over the next 2 years, the tumour recurred, and abdominal scans and subsequent biopsies confirmed the diagnosis of mesothelioma and showed metastatic disease around the liver, in the lymph nodes and in the peritoneum. He died at the age of 79 (54 months after diagnosis) years.

He was first exposed to asbestos 58 years prior to his diagnosis when he began a lengthy career as a Longshoreman in the shipping industry (likely mixed fibre types).

Case 6:

Case 6 is an African-American male aged 63 years at the time of diagnosis. Malignant mesothelioma (epithelioid type) was diagnosed on the right side of the scrotum after surgical biopsy and immunohistochemical staining. The tumour quickly metastasized to the kidney and lungs, and he died only 1 month after diagnosis.

He was first exposed to asbestos (fibre types unknown) 48 years prior to his diagnosis when he began work as a maintenance mechanic at an automobile manufacturing plant, which included exposure to asbestos products used in the physical plant of that facility. He held that job for

approximately 8 years. Further investigation into his work history is still ongoing.

Case 7:

Case 7 is a Caucasian male aged 51 years at the time of diagnosis. Malignant mesothelioma (epithelioid type) of the tunica vaginalis was discovered during a left-sided hydrocelectomy. The tunica vaginalis, tunica albuginea, spermatic cord, vas deferens, testicle and a section of scrotal skin were removed during surgery. The diagnosis was confirmed by a combination of cytology, surgical pathology and immunohistochemical staining. There has been no known recurrence to date (54 months from diagnosis).

He was first exposed to asbestos during his childhood, approximately 41 years prior to his diagnosis, when he began to assist his father on weekends with automotive brake and clutch work at his father's place of employment (most likely chrysotile-containing friction products with trace tremolite/actinolite). His own subsequent employment included 2 years working at a major military installation in a job that involved sweeping up the motor pool repair shop. He also worked for approximately 18–24 months as a truck driver, which sometimes included picking up, hauling and delivering asbestos-cement pipe (crocidolite- and chrysotile-containing). These runs would variously include picking up the pipe at the manufacturing plant or at a local supply house, unloading the pipe at his employer's warehouse, sweeping the truck after deliveries and delivering pipe to construction sites. He was also responsible for sweeping his employer's warehouse, including areas where the pipe was stored and where the pipe was cut by other workers using a power saw. He later worked for another trucking company for approximately 3 years and occasionally picked up, loaded and delivered

asbestos-cement pipe (crocidolite- and chrysotile-containing) and swept the truck.

Case 8:

Case 8 is a Caucasian male aged 51 years at the time of diagnosis. Malignant mesothelioma of the tunica vaginalis was diagnosed on the right side by surgical pathology and immunohistochemical staining. Two surgeries were performed that included the removal of the right testicle and spermatic cord, along with an epididymal mass, a scrotal scar and a right hydrocele sac. There has been no known recurrence to date (43 months from diagnosis).

He was first exposed to asbestos 31 years prior to diagnosis when he worked as a commercial and residential painter and was exposed to dust from a variety of construction activities, including the sanding of sheetrock (fibre types unknown but likely included chrysotile-containing taping compounds). He later worked a very short period of time as a contractor doing painting, sandblasting and cleaning work at petrochemical plants (fibre types unknown). He also had paraoccupational exposure to asbestos over many years doing automotive repair work to various vehicles owned by himself and by friends, including gaskets, brakes and clutches for motorcycles, cars and light trucks (likely chrysotile-containing with trace tremolite/actinolite).

Case 9:

Case 9 is a Caucasian male aged 65 years at the time of diagnosis. Malignant mesothelioma (epithelioid type) was diagnosed by surgical pathology and immunohistochemical staining. The tumour involved the right testicle, spermatic cord and 'peritesticular soft tissue'. Two years after the initial diagnosis and treatment, the tumour recurred on the left side and within 6 months had spread to the shaft of the penis, resulting in

additional surgery and requiring the removal of the left testicle and the penis itself. There has been no known recurrence since that time. He is now 39 months from his initial diagnosis.

He was first exposed to asbestos 49 years prior to his diagnosis when he first started working on his own automobiles, changing brakes, clutches and gaskets (likely chrysotile), which he continued to do for approximately 20 years. He also worked as a professional auto mechanic for 3 years where he had exposure to brakes, clutches and gaskets (likely chrysotile).

Table 1 contains summary data on all nine cases, including age at diagnosis, and latency from approximate first exposure to asbestos. The mean age at diagnosis for the nine cases was 59.7 years. The mean latency from first potential asbestos exposure to diagnosis was approximately 46.4 years.

Discussion

The tunica vaginalis is a mesothelium-lined sac that results from closure of the superior portion of the processus vaginalis, the channel through which the testes descend from the

Table 1 Subjects and exposures

Subject age at diagnosis (years)	Approximate latency from first known asbestos exposure (years)	Domestic or bystander exposures	Occupational exposures
1 (60)	53	Domestic (child): Removal of pipe insulation Home renovation products	Asbestos-cement pipe Bystander: Friction products
2 (70)	64	Domestic (child): Father and uncle were shipyard workers Home renovation products	Professional vehicle mechanic
3 (59)	48	Home renovation products	Building renovation products U.S. Navy Railroad
4 (44)	26		Scrapper and equipment operator at mining company Professional vehicle mechanic
5 (74)	58		Longshoreman in the shipping industry
6 (63)	48		Maintenance at factory
7 (51)	41	Vehicle repair	Asbestos-cement pipe
8 (51)	31	Vehicle repair	Sheetrock taping compound Petrochemical plant worker
9 (65)	49	Vehicle repair	Professional vehicle mechanic

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abdominal cavity to the scrotal sac during foetal life. In the event that the processus vaginalis does not close completely, then the tunica vaginalis remains in communication with the peritoneal cavity. Mesothelioma of the tunica vaginalis might thus be considered to be a variety of peritoneal mesothelioma where the peritoneal lining extends into the scrotal sac. This report describes nine cases of malignant mesothelioma of the tunica vaginalis testis in men with histories of asbestos exposure.

Asbestos exposure has previously been noted in individual cases reported in the literature and in subsets of larger case series.

The first clear confirmation of mesothelioma of the tunica vaginalis was reported by Barbera and Rubin⁸, who described a (probably benign) papillary subtype. The first report of asbestos exposure in a patient with malignant mesothelioma of the tunica vaginalis was described by Fligel and Kaneko, who reported a case in a 68-year-old asbestos worker with a later-diagnosed pleural mesothelioma⁵. The authors were unclear whether the pleural mesothelioma was a metastasis or a new primary because the family of the worker denied an autopsy request. Karunaharan⁹ reported one case in a 40-year-old asbestos worker. Huncharek et al.¹⁰ described a case in a 45-year-old former electrical power plant worker who reported asbestos exposure from insulation.

Jones et al.¹¹ described 11 cases in the largest reported single series. Only one had a history of asbestos exposure (9%), but information about the subjects' asbestos exposure was available in only five of the cases. In their review of the then-existing literature, they noted that the issue of asbestos exposure was specifically addressed in only 27 of the 64 reported cases, and of those 27 cases, 11 (41%) had a history of occupational asbestos exposure.

Plas et al.¹² reviewed the literature on mesothelioma of the tunica vaginalis testis and concluded that 34.2% of all reported cases had a positive history of asbestos exposure; however, the real prevalence may have been underestimated, because sufficient information was only given in 51.4% of the reported cases of malignant mesothelioma of the tunica vaginalis. The authors noted that 'exposure to asbestos or asbestos-containing materials remains the only established risk factor'.

Attanoos and Gibbs⁴ documented asbestos exposure in one of their three cases (33.3%) and found 'occasional' asbestos bodies in the lung tissue from that patient. While they believed that 'the link with asbestos exposure appears to be less strong than with pleural and peritoneal mesothelioma', they still concluded that '[t]he results of this study strongly suggest an additional association between asbestos exposure and localized primary ovarian and testicular mesothelioma'. Similar to Plas et al. they cautioned that 'it is possible that in a significant proportion of the reported cases where no asbestos history is documented, exposure had occurred in the past, but because of the long latency from initial exposure to asbestos and development of the tumour and death of the patient, exposure has been forgotten or not enquired into'.

Perez-Ordóñez and Srigley¹³ reported a history of asbestos exposure in three (23%) of their own series and noted that 'exposure to asbestos remains the only known risk factor for the development of [malignant mesothelioma] in the tunica vaginalis'. As with Plas et al. and Attanoos and Gibbs, these authors cautioned 'the true prevalence of asbestos exposure is probably underestimated because of insufficient clinical information'.

More recently, Schure et al. (2006)¹⁴ described three cases where none of

the patients reported a known history of asbestos exposure. As other authors have noted, however, a failure of a patient to report a history of exposure does not necessarily mean that no exposure has occurred: 'We do know that many asbestos exposed individuals do not know they have been exposed when they have actually been exposed in the workplace or in the community. Thus, a mesothelioma labelled as 'idiopathic' may be due to the fact that the victim's exposure to asbestos is unknown or documented'¹⁵.

Other investigators have reported the occurrence of this rare tumour in experimental animals, although the potential significance of their findings seemed to be lost, disregarded or misunderstood. Wagner et al.¹⁶ reported the occurrence of two mesotheliomas of the tunica vaginalis in their inhalation experiments with rats. They noted that their 'experiments do give some support to an association between asbestos exposure and tumours of the male genito-urinary system', although neither result was significant possibly due to the small control group. After including additional animals from other experiments in order to increase the size of the original control group, they concluded that 'there is no support for an association with tumours of the male genito-urinary system'. Suzuki and Kohyama¹⁷ induced peritoneal tumours of the tunica vaginalis testis in mice through the intraperitoneal administration of asbestos. They noted that 'there were certain 'favourite' sites of tumour production in the peritoneum', including 'the tunica vaginalis of the testis'. No other details about the number of such tumours or fibre type inducing the tumours were provided.

The men in this case series had all sought legal representation and were found to have had a variety of exposures to asbestos. Many were exposed as children during home renovation

work or to dust brought by relatives to their homes. Adult exposures included those from home renovation and construction work, vehicle maintenance, shipyard work and exposures at railroads. The pattern of exposures is similar to that described by Welch and colleagues¹⁸ in a case series of men with peritoneal mesothelioma and by Boffetta in a review of peritoneal mesothelioma in cohort studies¹⁹.

Conclusion

Nine American men diagnosed with malignant mesothelioma of the tunica vaginalis testis sought legal representation. All of the men had positive histories for occupational exposure to asbestos. Asbestos exposure should be considered an important risk factor for malignant mesothelioma of the tunica vaginalis testis.

Competing interests

Mr. Meisenkothen represents plaintiffs in asbestos litigation. Dr. Finkelstein has acted as a consultant for plaintiff's attorneys in asbestos litigation.

Consent

Written informed consent for publication of this case series was obtained from each living patient or from the next of kin for each deceased patient. A copy of the written consents is available for review by the Editor-in-Chief of this journal.

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