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Comprehensive description of adult-onset Still's disease after COVID-19 vaccination

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ABSTRACT

Cases of adult-onset Still's disease (AOSD) have been reported after COVID-19 vaccination. Here we provide a comprehensive description and analysis of all cases of AOSD reported in the literature and in pharmacovigilance databases through April 2022. Disproportionality analyses of pharmacovigilance data were performed in order to further explore the association between vaccination and AOSD. We included 159 patients, 144 from the World Health Organization pharmacovigilance database and 15 from the literature. Detailed clinical characteristics were described for the cases from the literature and from the French pharmacovigilance database (n = 9). The cases of AOSD after COVID-19 vaccination concerned women in 52.2% of cases. The median age was 43.4 years. More than 80% of AOSD reports occurred during the first three weeks and concerned mostly the BNT162b2 mRNA vaccine. We identified 14.5% of disease flare with a median time-to-onset of AOSD flare-up significantly shorter than for the new onset form. More than 90% patients received steroids. Although all cases were considered serious and required hospitalization, most cases presented a favorable outcome (67.1%) with a good response to corticosteroid therapy with a mean time to recovery of 7.2 days. Disproportionality analyses suggested that AOSD was associated with COVID-19 vaccines as well as other vaccines. AOSD was nearly five times more frequently reported with COVID-19 vaccines than with all other drugs. Clinicians should be informed about the potential risk of AOSD onset or flare following COVID vaccines and the importance of its early detection to optimize its management.

1. Introduction

In 2019, the global outbreak of SARS-CoV-2 infection, called COVID-19 disease, has motivated the extremely rapid development and approval of new vaccine platforms. The global pharmacovigilance systems have adapted their surveillance system to ensure enhanced surveillance of these new drugs [1,2]. This system has proven to be extremely effective in detecting safety signals [3,4] with a real-time evaluation of the benefit/risk balance. COVID-19 vaccination, like COVID-19 disease itself, induces an inflammatory reaction [5,6]. Unlike inactivated vaccine platforms, non-replicating viral vector (AstraZeneca, Janssen) and mRNA (Pfizer, Moderna) vaccine platforms have the ability to induce both cellular and humoral immune responses [7].

Adult-onset Still's disease (AOSD) is the adult form of systemic juvenile idiopathic arthritis which is a rare systemic inflammatory disorder that primarily affects young adults and mainly involve innate

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immune response. It corresponds to an aberrant inflammatory response whose precise etiology remains unknown. The role of an infectious (viral or bacterial) or non-infectious triggering factor on the ground of genetic susceptibility is generally accepted [8]. The diagnosis generally remains a diagnosis of exclusion based on nonspecific clinical criteria [9]. The Yamaguchi's criteria are the reference classification criteria set for this disease [10]. Currently, the precise pathogenesis of this disease and its optimal management are not consensually validated. Hemophagocytic lymphohistiocytosis (HLH) also known as macrophage activation syndrome (MAS) is considered as the most serious life-threatening complication of AOSD. Cases of AOSD and HLH have been reported after COVID-19 disease [5,11,12].

Among the unexpected cases of adverse drug reaction after COVID-19 vaccination, some cases of AOSD have been described in literature and reported to the global pharmacovigilance system.

The objective of this study is to present the most complete description of all cases of AOSD reported after vaccination with COVID-19 both in literature and in pharmacovigilance databases in order to improve knowledge of the development of this disease after vaccination. We hence provided a clinical description containing the main clinical symptoms, the time to onset after vaccination, the different therapeutic strategies adopted and the evolution after treatment. We also sought to better characterize the risk of occurrence of this rare but serious adverse event after COVID-19 vaccination.

2. Methods

We collected all cases reported in the World Health Organization's pharmacovigilance database, VigiBase until April 2022 and the cases of AOSD after COVID-19 vaccination described in literature. VigiBase is a global database collecting more than 23 million adverse drug reaction reports from over 150 countries worldwide. The literature review has been performed using PubMed with the terms 'Adult-onset Still's disease' and 'COVID-19 vaccine'. For the VigiBase query, we selected all de-duplicated individual case safety report (ICSR) of adult-onset Still's disease (Preferred Term of the Medical Dictionary of Regulatory Activities, MedDRA 25.0) reported with all COVID-19 vaccines by using the narrow Standardized Drug Groupings (SDG) 'Vaccines for COVID-19'. ICSRs contain patient characteristics such as age and sex, reporter qualification, data about reported drugs including dose, date of administration and occasionally vaccine rank, and all adverse drug reactions (ADRs) coded according to MedDRA, with date of occurrence and evolution. The exclusion criteria were age less than 18 and cases assessed as potential duplicate because of same age, sex, country, vaccination date, same vaccine with identical batch number and same time to onset of AOSD. Cases of AOSD onset more than one year after COVID-19 vaccination have been excluded [13]. We have considered as AOSD flares the cases in whom the terms 'disease worsening' or 'flare' were co-reported or where a history of AOSD was mentioned. All others cases were considered as new AOSD onset.

We performed disproportionality analyses using the case/non-case method which allows to identify a disproportionate reporting, i.e. a higher than expected number of adverse reaction reports compared to other reactions recorded in the database by calculating Reporting Odds Ratios (ROR) [14]. Cases were all reports of AOSD reported and non-cases were all other serious drug-related adverse reactions recorded in this global database. Exposure to COVID-19 vaccines (BNT162b2, mRNA-1273, ChAdOx1 nCoV-19, JNJ 78436735) was compared between cases and non-cases to all other drugs and, in a secondary analysis, to all other vaccines. A signal of disproportionate reporting was considered if the 95% confidence interval (95% CI) lower limit of the ROR exceeded 1.

Cases extracted for the French Pharmacovigilance Database (FPVD) are included in the WHO database but also contain, beyond the data available in VigiBase, the detailed clinical course of the ADR, the steps of diagnosis and the management of the ADR. These cases allowed us to perform a combined descriptive and comparative analysis with the cases extracted from literature regarding type of vaccine, time to onset after vaccination, AOSD classification criteria of Yamaguchi, ferritin level, management and evolution with time to resolution. More commonly used than Fautrel criteria [15], Yamaguchi criteria are sensitive in the diagnosis of AOSD if the patient fulfill five criteria with at least two major and do not present exclusion criteria [10].

3. Results

After excluding 4 duplicates and 9 cases occurring in children (<18 years), we selected 145 cases of AOSD associated with COVID-19 vaccine from the query conducted in VigiBase on April 20, 2022. No cases were excluded based on an inconclusive time to onset (TTO). The literature review reported 15 cases [16–27], of which only one was also identified in VigiBase (see Fig. 1). In total, 159 reports have been included in the main analysis and are presented in Table 1. Of the 22 countries providing contributing data, the United States of America contributed for nearly half of reports with a lower completeness score. Reporters were mostly health professionals. For all selected reports, the COVID-19 vaccine was considered as the single suspected drug for the onset of AOSD. AOSD cases showed a median age of 43 years and a slight female predisposition. The suspected vaccines were mRNA-based COVID-19 vaccines in 72.3% of the cases. Among the 159 cases, we identified 23 (14.5%) flare of the disease. Among the cases with available information on dose rank (n = 45), AOSD occurred for nearly two third after the first dose. For three cases (6.7%), the first two doses have been considered suspect: in two cases, there was a single event after the second dose, but the involvement of the first dose have not been excluded; the third case concerns a patient who presented a flare-up of his disease after each of the two doses of vaccine. Time to onset ranged from 0 days to 6 months with a median of 8 days (IQR 3–18 days). AOSD occurred in more than 80% during the first three weeks after vaccination. Most of the cases (86.8%) have been reported as serious including 18 (11.3%) life-threatening conditions and two (1.2%) fatal outcomes. Co-reported terms such as 'myocarditis', 'pericarditis', 'myopericarditis' and 'hemophagocytic lymphohistiocytosis' (HLH) have been screened both in VigiBase and literature data. We identified 19 (11.9%) cases with a co-reported cardiac affection and 11 (6.9%) cases with HLH of whom 3 presented also a myocarditis. Among patients with available evolution (n = 83), 67.1% have recovered or were recovering from the event.

Disproportionality analyses showed that, among all serious ADR reported in the WHO pharmacovigilance database, AOSD was nearly five times more frequently reported with COVID-19 vaccines than with all other drugs (ROR = 4.96; 95%CI: 4.11–5.99). However, AOSD was not more reported with COVID-19 vaccines than with all others vaccines (ROR = 0.91; 95%CI: 0.67–1.23). As VigiBase data are clinically limited, more qualitative clinical data extracted from the literature cases (n = 15) and the French pharmacovigilance database cases (n = 9) are presented in Table 2. Detailed individual clinical features of these cases are available in Supplementary Table. Among these 24 cases, 22 met Yamaguchi criteria and 20 Fautrel criteria, all literature reports and 7 extracted from FPVD because of some missing data. Twenty-three cases (95.8%) presented ≥3 major criteria and 14 cases (58.3%) presented all major criteria, 16 with hyperleukocytosis and only one case that did not showed typical skin rash. We did not find any exclusion criteria available. Concerning minor criteria, we have found at least three minor criteria for 15 cases (62.5%) (Table 2). Hyperferritinemia was reported in 20 cases (83.3%). In the clinical course of AOSD some complications may occur. Three patients (12.5%) have presented macrophage activation syndrome with very high levels of ferritinemia (≥20,000 ng/ml). Less frequent manifestations of AOSD have also been described that notably include cardiac involvement. Noteworthy, five cases of AOSD (20.8%) have been reported with associated myocarditis or myopericarditis with high troponin level.

Regarding treatment, more than 90% patients have received

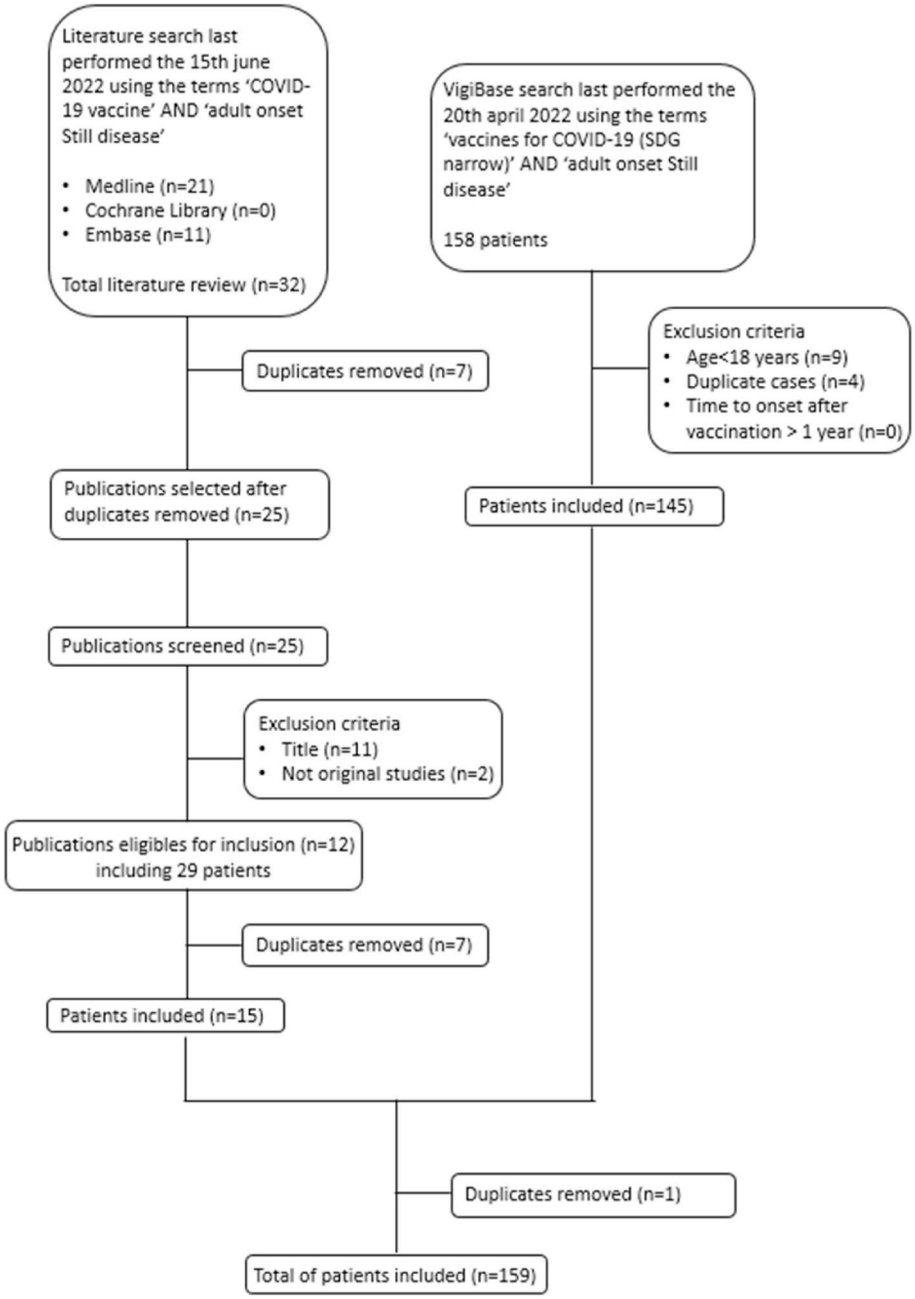

Fig. 1. Flow diagram of the selection of cases from literature and pharmacovigilance databases.

steroids, fourteen by intravenous route (pulses). Fifteen of them (62.5%) required additional therapy: anti-IL-1 or anti-IL-6 in 45.8% of cases (anakinra n = 4 and tocilizumab n = 7), methotrexate in 4 cases and IVIg in 3 cases. The outcomes have been generally favorable with a rapid regression of symptoms. IL-6 levels were previously assessed in only 2 of the 11 patients who received treatment with anakinra or tocilizumab. The level was abnormally high in only 1 patient [16,17]. For the thirteen documented cases the mean and the median time to recovery have been 7.2 days (SD 8.6 days) and 4 days respectively. Interestingly the median TTO of flare-up AOSD (5 days) was significantly shorter than for the new onset form (10 days) (Wilcoxon - Mann Whitney test, $p = 0.02$). However, the outcomes of the flare-up cases were also rapidly favorable after treatment. To our knowledge, there is no recommendation for AOSD flare-up management. In this study, when information was available (n = 5), treatment was always based on corticosteroids and anti-IL-1 or anti-IL-6 (see more details in Supplementary table). Most of them have previously received steroids and methotrexate. One patient had been stabilized for only 3 months with anakinra and prednisolone, one had been in remission for 1 year with tocilizumab, one for 14 years with weekly etanercept and low dose of prednisone, and one for 21 years with methotrexate.

4. Discussion

As with any new drug, rare adverse events cannot be detected during clinical trials due to the limited number of patients tested and the selection criteria. Therefore, in the post-marketing period, pharmacovigilance remains an essential system for consolidating drug safety profiles and detecting previously unknown and rare ADRs [28]. Herein, based on literature review and analyses of pharmacovigilance databases, we present the largest series of AOSD cases that have been reported with COVID-19 vaccination. We describe 159 cases with a median age at

Table 1
Characteristics of adult-onset Still's diseases reported after COVID-19 vaccination in pharmacovigilance databases (n = 144) and in the literature (n = 15).

Characteristics	Cases (n = 159)
Age (years), median (P25–P75) (n = 110)	43.4 (33.0–53.0)
Gender, n (%)	
Male	76 (47.8)
Female	83 (52.2)
Reporter type, n (%)	
Health professional	53 (67.9)
Other	25 (32.1)
Unknown	81
Reporting year, n (%)	111 (69.8)
2021	
2022	48 (30.2)
Reporting countries, n (%) – Mean CS (/1)[a]	
United States of America	79 (49.7) – 0.30
Italy	14 (8.8) – 0.86
France[b]	10 (6.3) – 0.92
Germany	10 (6.3) – 0.90
United Kingdom	8 (5.0) – 0.66
Australia	8 (5.0) – 0.32
Others (16 countries)	30 (18.9) – 0.70
Reaction reported, n (%)[c]	
New onset of AOSD	136 (85.5)
AOSD flare	23 (14.5)
COVID-19 vaccine	
mRNA vaccines	
BNT162b2	102 (64.2)
mRNA-1273	13 (8.2)
Adenovirus vaccines	
ChAdOx1 nCoV-19	37 (23.2)
JNJ 78436735	7 (4.4)
Vaccinal dose, n (%)[d]	
D1	28 (62.2)
D2	11 (24.4)
D3	3 (6.7)
D1+D2	3 (6.7)
Unknown	114
Single suspect	159 (100.0)
Time to onset, median (IQR)	8 (3–18)
Time to onset, n (%)	
<1 month after vaccination	
J0-J7	56 (46.3)
J8-J14	29 (24.0)
J15-J21	12 (9.9)
J22-J28	1 (0.8)
≥1 month after vaccination	
1 month	10 (8.3)
2 months	7 (5.8)
≥3 months	6 (4.9)
Unknown	38
Serious, n (%)	138 (86.8)
Seriousness criteria, n (%)	
Caused/prolonged hospitalization	125 (78.6)
Other medically important condition	18 (11.3)
Life threatening	18 (11.3)
Disabling/incapacitating	8 (5.0)
Death	2 (1.2)
Co-reported MedDRA, n (%)	
Myocarditis	10 (5.7)
Pericarditis	8 (5.0)
Myopericarditis	1 (0.6)
Hemophagocytic lymphohistiocytosis	11 (6.9))
Evolution, n (%)	
Recovered	34 (41.5)
Recovering	21 (25.6)
Recovered with sequelae	2 (2.4)
Not recovered	24 (29.3)
Died	2 (1.3)
Unknown	76

[a] CS: completeness score.
[b] Case reported by the manufacturer and not recorded in the FPVD.
[c] Relapse of preexisting Still's disease assessed with 'disease worsening' or 'flare of the disease' co-reported terms.
[d] Vaccinal dose: D1: dose 1, Dn: Dose n.

Table 2
Clinical features of adult-onset Still's diseases reported in literature and in the French Pharmacovigilance database from January 2021 to July 2022.

Clinical features	Cases (n = 24 = 15 + 9)
Age (years), median (P25–P75)	37.5 (30.5–47.5)
Gender, n (%)	
Male	10 (41.7)
Female	14 (58.3)
Reaction reported, n (%)	
New onset of Still's disease	15 (62.5)
Relapse of a preexisting Still's disease	6 (25.0)
Time to onset (days), median (IQR) (n = 21)	7 (5–13)
New onset cases	10 (6–17)
Flare Up cases	5 (2.5–6.75)
Yamaguchi criteria, n (%)	
Major	
Fever ≥39 °C, for 1 week or more	22 (91.7)
Arthralgia for 2 weeks or more	24 (100.0)
Typical skin rash	23 (95.8)
Hyperleukocytosis	16 (66.7)
Minor	
Pharyngitis or sore throat	19 (79.2)
Lymphadenopathy or splenomegaly	10 (41.7)
Elevated transaminases	13 (54.2)
Absence of rheumatoid factor or antinuclear antibodies	17 (70.8)
Hyperferritinemia reported, n (%)	20 (83.3)
Ferritin level (ng/ml), min-max	404-136,680
HLH, n (%)	3 (12.5)
Cardiac abnormalities, n (%)	7 (29.2)
Myocarditis	4 (16.7)
Pericarditis	1 (4.2)
Myopericarditis	1 (4.2)
Cardiac biomarkers	4 (16.7)
Treatment	15 (62.5)
Corticosteroid therapy, n (%)	15 (62.5)
≥1 mg/kg	4 (16.7)
Additional therapy	7 (29.2)
Anti-IL-1	4 (16.7)
Anti-IL-6	3 (12.5)
MTX	
IVIg	
Time to recovery (days), median (IQR) (n = 13)	4 (3–7)

onset of 43 years. A majority of cases concerned mRNA-based COVID-19 vaccines. We identified a significant and probably underestimated proportion of disease flares in patients with preexistent AOSD consistent with a review of all autoimmune or inflammatory diseases reported after COVID-19 infection or vaccination [29]. AOSD after vaccination occurred mostly after the first dose and generally during the first three weeks following vaccination. Interestingly, our study shows a significantly shorter time to onset for flares than for new onset forms. Hyperferritinemia was present in more than 80% of the AOSD cases described in our study. Ferritin levels were shown to be significantly higher in patients with severe Covid-19, questioning its role in the pathogenesis of auto-inflammatory and autoimmune diseases [30]. Fautrel criteria have also been reviewed in all these cases even if the glycosylated ferritin level is often missing. Despite the seriousness of this ADR, few cases reported a fatal outcome. A favorable evolution was generally observed after treatment, mainly based on steroid therapy. Even if a definite causal link between AOSD and COVID-19 vaccination could not be asserted, our disproportionality analyses suggested that COVID-19 vaccines could increase the risk of AOSD. However, AOSD was not more frequently reported with COVID-19 vaccines than with other vaccines. It has been shown that vaccination and natural infections can cause new onset or flare of autoimmune and inflammatory diseases [6,29,31,32]. Furthermore, in the last decade, Shoenfeld and colleagues introduced the notion of adjuvant-induced autoimmune (auto-inflammatory) syndrome (ASIA) [33]. The role of adjuvants, particularly aluminum, is to increase the immune response and its duration [34]. Although Covid-19 mRNA-based or non-replicating viral vector vaccines do not contain adjuvants, autoimmune and autoinflammatory diseases

have been described with these vaccines [35]. AOSD is a rare multisystem auto-inflammatory disorder characterized by cardinal symptoms, (skin rash, fever, hyperleukocytosis and arthritis or arthralgia) and other manifestations, making clinical expression heterogeneous and diagnosis sometimes difficult. Although poorly understood the pathogenesis is related to inflammation mediated by activation of the innate immune system with abnormal production of proinflammatory cytokines, including interleukin (IL)-1β, IL-18, IL-6, and tumor necrosis factor-α (TNF-α) after a potential trigger factor on a predispose genetic background [8,36,37]. Vaccines have also been already proposed as potential triggers of AOSD notably influenza, hepatitis A and B and pneumococcal vaccines have been already suggested [38–41]. AOSD has also been described after COVID-19 infection [11,12]. Even if the mechanism by which COVID-19 infection or vaccination could trigger AOSD is not completely understood some explanations have been proposed, notably anti-idiotype immune response [42,43]. Among them, molecular mimicry which relies on immune cross reactivity due to structural homology between pathogens and self-proteins [31]. It has been shown that the SARS-CoV-2 spike protein can be recognized as a pathogen-associated molecular patterns (PAMP) by pattern recognition receptors (PRRs), such as Toll-like receptors (TLRs), NOD-like receptors (NLRs), and RIG-I like receptors (RLRs) [44]. TLRs thereby activating the early stages of the innate immune response with the expression of inflammatory mediators [45]. Both mRNA and adenovirus vaccines could stimulate innate immunity through PRR [46]. This recognition subsequently causes the release of proinflammatory cytokines and activates an adaptive immune response.

The multiplication of post-vaccine AOSD cases reported in the literature and to the pharmacovigilance systems has underlined the importance to explore this potential signal. We carried out an exhaustive review of cases reported worldwide. In line with this, we performed a systematic review of all PubMed case reports, together with an analysis of pharmacovigilance data recorded in French and global databases. The BNT162b2 vaccine was the most represented (72.3%), reflecting its widespread diffusion on a global scale. We selected and reviewed 159 cases. Among them, the modest over-representation of women (52.2%) is not sufficient to suggest female predominance. A similar gender distribution is generally found in idiopathic case series where females represent 45–53% of patients [37,47]. In terms of age, in our dataset and according to the literature, adults between 18 and 65 years of age were more frequently prone to AOSD in contrast to the elderly.

A part of our analysis focused on identifying cases reporting a worsening or flare-up of the disease. We identified 23 cases (14.5%) likely to be relapses of the disease reinforcing previous reports of flares of pre-existing autoimmune diseases after COVID-19 vaccination [35]. Our study also shows that the disease was more often reported after the first dose of vaccine (62.2%) and during the first two weeks after vaccination (70.3%) which is consistent with average time-to-onset of other autoimmune or autoinflammatory conditions previously described after COVID-19 vaccination (from 8 days to 3 weeks) [48]. Information about rechallenge is extremely rare but one case from the literature describes an outbreak of the disease after each of the first two doses highlighting the potential link between vaccination and flares [16].

In a recent review of ten case reports study, Winichakoon et al. [45] showed that the clinical features of AOSD following COVID-19 vaccination were mainly consistent with large series of idiopathic AOSD. In addition to the description of general characteristics in 159 cases, our study adds the well-documented reports of the french pharmacovigilance database allowing us to specify the clinical features on 24 reports in total.

HLH is a potential life-threatening complication of AOSD that shares common pathophysiological pathways. In the same way than other multisystemic inflammatory syndromes, HLH have been described after COVID-19 disease and COVID-19 vaccination. We observed a similar occurrence of HLH in our case review after COVID-19 vaccination than reported in idiopathic AOSD [8,49].

Winichakoon and colleagues suggested a greater proportion of cardiac involvement in AOSD after COVID-19 vaccination. COVID-19 disease, like other viral infections, is well known to induce myocarditis with a higher frequency than the myocarditis or pericarditis occurring after COVID-19 vaccination [50,51]. The inflammatory mechanisms involved in the development of these adverse events might overlap. It is also known that cardiac manifestations are frequent in AOSD and sometimes inaugural [52]. In the present study, we identified 19 cases (11.9%) with myocarditis, pericarditis or myopericarditis co-reported with AOSD. This proportion was not increased compared to idiopathic AOSD case series [52]. Thus, it is difficult to conclude whether cardiac events are directly related to AOSD or are another adverse effect of vaccination independent of AOSD.

Interestingly, the TTO was significantly shorter in episodes identified as flares compared to those identified as inaugural disease. This result suggests that COVID-19 vaccines could potentially trigger AOSD flares.

The majority of our cases presented a favorable outcome with a good response to steroid therapy. Indeed, corticosteroid therapy remained the main reported treatment in our series. However, additional therapy was required in more than half of the cases and biologics in more than 45%. Methotrexate was administered in 16.7% mainly as a steroid-sparing strategy [53]. Anti-interleukin therapies have been also used to treat serious COVID-19 disease [54]. Even in absence of management guidelines of AOSD patients, a dichotomous view of this disease have been proposed with different cytokine profiles and responses to biologic treatments [55].

Our work included a selection of ADR reports associated with COVID-19 vaccines up to April 2022. Although the VigiBase query was conducted using the Standardized Drug Groupings 'Covid 19 vaccines', only vaccines approved in the countries with the highest contribution to the WHO database were found in the results. The systematic review of the WHO database in combination with the literature therefore provided a complete collection of available cases of AOSD occurring after COVID-19 vaccination worldwide. However, this comprehensive study presents some limitations mostly inherent to the underreporting of ADR. Thus, reporting rate cannot be interpreted as real incidence. In our series, the low rate of cases both described in the literature and reported in VigiBase further illustrates this under-reporting phenomenon and the rarity of AOSD in general. We evaluated the association between AOSD and COVID-19 vaccines through disproportionality analyses. Based on reported data only, this method cannot assert causality and measure real risks. However, in many examples, disproportionate reporting has proven to be useful to identify drug-related risks [56]. Another limitation of VigiBase analysis is missing data, such as past medical history, complete work-up, ferritin level, co-reported affections and management strategies. The outcomes are poorly documented in the international ICSR. This lack of clinical outcome information can be partly explained by an early reporting but also by the long and complex management of these patients, maybe lost to follow-up. Nevertheless, in our study, the high quality and completeness of the French reports allowed us to assess the missing clinical features with the same criteria as the cases in the literature. Our study thus combined the power of the largest data set with the precision of a qualitative sample highlighting the complementarity of these different post-authorization surveillance methods.

5. Conclusion

Our study suggests a role of COVID-19 vaccines in occurrence of AOSD as already hypothesized with other vaccines. The diagnosis of AOSD remains a diagnosis of exclusion difficult to establish with a risk of underestimation of the real incidence. Nevertheless, the small number of reported cases of AOSD compared to the number of people vaccinated does not question the benefit of vaccination even concerning healthy people. Further studies, such as pharmacoepidemiological studies, will be needed to compare the occurrence of AOSD after vaccination to the

natural incidence of this disease and to its incidence after COVID-19 disease. Physicians should be aware of this very rare condition in patients who presented prolonged fever shortly after COVID-19 vaccination. Prompt recognition of AOSD is essential for proper early management in order to avoid potential serious complications.

Author contribution

PP and VB equally contributed to the design of the study, the acquisition and interpretation of data, drafting, revising and approving the manuscript; ATJM and JLF contributed to the conception of the work, revising, criticizing and approving the manuscript; All authors agree to be accountable for all the aspects of the work (notably accuracy/integrity of data)

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Declaration of competing interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

Data availability

Data will be made available on request.

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Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.jaut.2022.102980.

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